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Conceptualisation of Patient- and Family-Centred Care for Young Adults with Hearing Loss and their Family Members

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ABSTRACT

The period of life between 16 and 25 years of age is an important time of change and development, with many young people leaving home, moving into tertiary education, establishing careers, or beginning families of their own. In Australia, more than 8000 young adults are living with a hearing loss requiring hearing rehabilitation. These hearing losses have the potential to have significant and far-reaching consequences for the social, emotional, educational, and vocational lives of these young adults, and it is currently unclear what these effects might be, or whether hearing rehabilitation is reducing them. In addition, while it is known that the effects of chronic health conditions including hearing loss can be effectively reduced by the application of patient- and family-centred care (PFCC), it is not currently clear what constitutes PFCC in this population.

This thesis aimed to explore the lived experiences of young adults with hearing loss and to identify the nature of high-quality PFCC among them and their family members. Beginning with two systematic reviews of previously-published research addressing PFCC among young adults with other chronic health conditions, the likely primary elements of PFCC were identified, as well as evidence suggesting that attempts to implement these elements are likely to result in improved outcomes for young adults with chronic health conditions.

Following this preparatory work, a survey was conducted to explore the demographics of young adults with hearing loss in Australia, and compare these to those of other young adults in Australia. Significantly poorer life satisfaction outcomes were found for young adults with hearing loss, although a correlation between life satisfaction and the patient-centredness of hearing (re)habilitation was identified, supporting the importance of patient-centred intervention in this population.

Following this quantitative work, a group of young adults with hearing loss were interviewed, and asked about their experiences of hearing (re)habilitation, with a view towards identifying the major components of high-quality hearing (re)habilitation. Similar to other work in this field, the importance of a strong therapeutic relationship was identified, supported by hearing services that provided a service that was desired and valued, who were experts in hearing loss and its (re)habilitation, and who allowed young people control over their care and related decisions.

Finally, a group of mothers of young adult children were interviewed, who told their stories of hearing (re)habilitation for their children. These narratives, beginning at diagnosis and stretching through to the present day (in which they were largely uninvolved in the day-to-day of hearing (re)habilitation) reflected a shared quest among these mothers to produce independent, successful children who were able to self-advocate and self-manage. These results underscored the importance of being aware not only of the dynamics of rehabilitation within the clinic room but also how those dynamics may have grown and developed over time.

This research has shown that hearing loss is associated with significant impacts on young people, and that these impacts are not fully remediated by hearing (re)habilitation as it is currently delivered. However, it also presents a model of care that may assist hearing services to provide desirable PFCC for their young adult patients and their families in the future. This work strongly supports the incorporation of young people and families into decision-making and service design, as well as continued research into the needs and desires of young people living with hearing loss.

DECLARATION BY AUTHOR

This thesis is composed of my original work, and contains no material previously published or written by another person except where due reference has been made in the text. I have clearly stated the contribution by others to jointly-authored works that I have included in my thesis.

I have clearly stated the contribution of others to my thesis as a whole, including statistical assistance, survey design, data analysis, significant technical procedures, professional editorial advice, financial support and any other original research work used or reported in my thesis. The content of my thesis is the result of work I have carried out since the commencement of my higher degree by research candidature and does not include a substantial part of work that has been submitted to qualify for the award of any other degree or diploma in any university or other tertiary institution. I have clearly stated which parts of my thesis, if any, have been submitted to qualify for another award.

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PUBLICATIONS INCLUDED IN THIS THESIS

The following refereed journal publications are incorporated into this thesis.

1. Allen, D., Scarinci, N., & Hickson, L. (2018). The Nature of Patient- and Family-Centred Care for Young Adults Living with Chronic Disease and their Family Members: A Systematic Review. International Journal of Integrated Care, 18(14). doi:10.5334/ijic.3110/

   Incorporated as Chapter 2.

SUBMITTED MANUSCRIPTS INCLUDED IN THIS THESIS

The following papers submitted for peer review by journals are incorporated into this thesis.

1. Allen, D., Scarinci, N., & Hickson, L. (in press). Effects of Patient- and Family-Centred Care for Young Adults with Chronic Health Conditions and their Family Members: A Systematic Review. *Emerging Adulthood*.

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OTHER PUBLICATIONS DURING CANDIDATURE

No other publications.

CONTRIBUTIONS BY OTHERS TO THE THESIS

The PhD candidate was primarily responsible for the conceptualisation and design of each of the studies contained within this thesis, as well as gaining ethical approval, participant recruitment, data collection, analysis, and interpretation, and manuscript preparation. However, significant contributions have been made by the following people:

Doctor Nerina Scarinci and Professor Louise Hickson had substantial input into the conceptualisation and design of each study, the analysis and interpretation of the data, and editing of the manuscript.

Alison King had significant input into the design, data analysis, and editing of the manuscript of the study presented in chapter 4.

Professor Robert Cowan had significant input into the editing of manuscripts of all studies.

To the best of my knowledge and belief, all persons who made a contribution to this work have been acknowledged consistent with The University of Queensland policy, particularly PPL 4.20.04 Authorship and its associated codes, policies, and procedures.

STATEMENT OF PARTS OF THE THESIS SUBMITTED TO QUALIFY FOR THE AWARD OF ANOTHER DEGREE

No works submitted towards another degree have been included in this thesis.

RESEARCH INVOLVING HUMAN OR ANIMAL SUBJECTS

The portions of this research involving human participants were approved by The University of Queensland Human Research Ethics Committee A (Approval number 2016001270), the Australian Hearing Human Research Ethics Committee (AHHREC), and the Hear and Say Research and Ethical Advisory Committee. No certificate was issued by the AHHREC.

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KEYWORDS

Young adults, emerging adults, hearing loss, chronic disease, hearing rehabilitation, patient-centred care, patient- and family-centred care

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DEDICATIONS

To Mum, who taught me that the world can be better if we make it;

To David, who never stopped believing in me;

To Jack, who never let me say “I can’t”;

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To all my participants, who shared so much of themselves:

This one’s for you.

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LIST OF ABBREVIATIONS USED IN THE THESIS

ACT	Acceptance and Commitment Therapy
CASP	Critical Appraisal Skills Program
FCC	Family-Centred Care
HAUQ	Hearing Aid Users Questionnaire
HRQoL	Health-Related Quality of Life
ICD-10	International Classification of Diseases, Tenth Revision
ICD-10-CM	International Classification of Diseases, Tenth Revision, Clinical Modification
ICF	International Classification of Functioning, Disability, and Health
LSAY	Longitudinal Surveys of Australian Youth
MeSH	Medical Subject Headings
MPOC-A	Measure of Processes of Care for Adults
PCC	Patient-Centred Care
PFCC	Patient- and Family-Centred Care
PFCI	Patient- and Family-Centred Interventions
SF-36	36-Item Short Form Health Survey
QARI	Qualitative Assessment and Review Instrument
WHO	World Health Organization

CHAPTER 1 – BACKGROUND AND METHODOLOGY

Introduction

Hearing loss is one of the most common chronic health conditions affecting people worldwide, and patient- and family-centred care (PFCC) has been shown to be an effective approach to improving the lives of people with hearing loss. A significant number of emerging adults (between the ages of 16 and 25) are currently living with hearing loss, and the changes undergone over the course of this period of life can complicate the delivery of appropriately patient- and family-centred hearing (re)habilitation.

In this chapter, general background information on chronic health conditions and the nature of PFCC is presented, along with evidence for the status of hearing loss as a chronic health condition, and the importance of the use of PFCC in the treatment of chronic health conditions. Finally, the particular challenges of treating emerging adults in general, and those with hearing loss in particular, are presented, leading to the aims of this thesis as a whole.

What are chronic health conditions?

Chronic health conditions like hearing loss are negative states of health that persist over time and have complex effects on the person and their healthcare (Australian Institute of Health and Welfare, 2014, p. 94). They are characterised by long durations (more than three months), multifactorial causes and aetiologies, and changes in the nature and severity of symptoms over time. They may affect a wide variety of bodily systems, including the cardiovascular system, respiratory system, nervous system, and sensory organs (World Health Organisation, 2005). Importantly, chronic diseases do not have a definitive cure. Rather, the focus of medical management is on managing and slowing the progress and symptoms (Martin, 2007).

Most patients with chronic health conditions do not receive high-quality primary care, due to the increasing demands of practitioners' time and the difficulty of maintaining

continuity of care (Grumbach & Bodenheimer, 2002). While technological and organisational methods can support patients and improve care delivery, they can be complex for practitioners to implement (Bodenheimer, 2002a). In addition, as many chronic health conditions share risk factors, many patients live with multiple comorbidities across bodily systems and with varying treatment requirements. These "discordant comorbidities" tend to lead to reduced treatment effectiveness (Aung et al., 2015), both by directly affecting the patient's physical, mental, and emotional capability, and by demanding already limited clinical resources such that conditions that have a significant effect on quality of life are under-addressed (Starfield et al., 2003).

To reduce the impact on clinical resources, patients with chronic health conditions may be encouraged to self-manage, with responsibility for clinical tasks that may be readily performed outside the clinic being passed on to the patient and their family (Bodenheimer, 2002b). While this transfer of power and responsibility can reduce the need for intensive practitioner involvement, patients still see their practitioner as an important source of information and guidance through their care journey (Rademakers, Delnoij, Nijman, & de Boer, 2012), as well as a source of medical expertise separate from the patient's experiential expertise (Bodenheimer, 2002b).

The extended nature of management of chronic health conditions requires interaction between a patient and their healthcare team on an ongoing basis, potentially for many years. In addition, often several different healthcare providers, other professionals, and family members may be involved in delivering support and care to the patient, requiring detailed communication between them (World Health Organisation, 2005, p. 96). This network of people involved in supporting a person with a chronic health condition may also change over the lifespan. For example, a person with a profound chronic impairment may require support

from teachers and aides during the school years, occupational therapists during their working life, and nursing staff in older age.

In these complex management situations, patients may feel that they become experts in their own conditions (Kramer, 2005), and may gain significant benefit from managing much of the treatment process themselves rather than ongoing treatment being performed by a healthcare professional (Lorig, Sobel, Ritter, Laurent, & Hobbs, 2001). This empowerment of patients to manage their health conditions themselves can improve treatment efficacy (as treatment can be applied on an ongoing basis rather than only when the practitioner is available), reduce burden on the health system, and give the patient a sense that they are engaged with and in control of their care rather than being treated at the convenience of their practitioner (Bodenheimer, 2002b; World Health Organisation, 2011, p. 65).

However, patients will only be able to self-manage effectively when they are physically, mentally, and emotionally able to effectively apply the interventions in day to day life (Bodenheimer, 2002b). As a result, medical management of a chronic health condition needs to be integrated with the patient's lifestyle, social structures, abilities, and competencies (World Health Organisation, 2005, p. 35). . It is vital that practitioners ensure that the patient has the information, skills, and resources that they need in order to self-manage, and that they engage their family and other support structures in a patient- and family-centred manner.

What is Patient- and Family-Centred Care?

Patient-centred care (PCC) is care that puts the control for clinical encounters in the hands of the patient, rather than in the hands of the practitioner (Stewart et al., 2014, p. 7). It focuses on involvement of the patient in the healthcare process, addressing their needs specifically and allowing them to participate in and control decision making (Kitson, Marshall, Bassett, & Zeitz, 2013). As a result of this, it acknowledges that the patient is a

person, with individual needs and experiences of disease that are separate from the disease process itself: that is, that two people with similar or identical disease progression may nonetheless have significantly different experiences of illness and disability (Mead & Bower, 2000).

Importantly, patient-centred approaches often involve a redefinition of the nature of illness itself. Rather than the medical model of disease as a breakdown in a body system, patient-centred approaches to care use a biopsychosocial model of disease and illness. In these models of disease, a complex network of interactions between changes in body functions caused by the health condition, the person's ability to perform tasks, and their ability to engage in situations in their life all contribute to their experience of disease. These factors are further all modified by the person's personal, interpersonal, and environmental context (World Health Organisation, 2002). The inclusion of personal factors in this model helps to explain, for example, why some patients may find the exclusion of biological causes of disease reassuring, while for others it is a source of distress (Mead & Bower, 2000).

Fundamental to the patient-centred model of healthcare delivery is the development of a relationship between the patient and practitioner (Kitson et al., 2013), or "therapeutic alliance" (Mead & Bower, 2000). This alliance facilitates open and honest communication between patient and practitioner, allowing practitioners to gain a more integrated understanding of the patient's situation and experience, enhancing their delivery of services (Stewart et al., 2014, pp. 143-147). The practitioner gives the patient the information and power that they need to make decisions about their care, rather than acting as a "gatekeeper" to information and treatment.

Among children and younger people, parents and other family members may be the primary agents of decision-making and the implementation of treatment. In these cases, a purely patient-centred approach may not be appropriate, particularly if the patient does not

have the cognitive capacity to participate fully in independent shared decision-making and in the implementation of those decisions. In these cases, a Family-centred care (FCC) model may be more appropriate, which treats the family as the recipient of attention and the primary actor in the healthcare process (Epley, Summers, & Turnbull, 2010). FCC prioritises the family as the most stable part of a child's life, around which healthcare systems and practitioners may change (Shelton & Stepanek, 1994). The family is considered not only the primary source of knowledge about the child's health and functioning, they are also the most important and consistent delivery mechanism for healthcare interventions to the child. It assumes that parents are experts on their children, and want positive outcomes for them (Rosenbaum, King, Law, King, & Evans, 2009).

As a result, FCC holds that integrated, collaborative action between healthcare providers and families, with full disclosure of all relevant information in both directions, is the most effective way to achieve healthcare outcomes for children (Shelton & Stepanek, 1994). This interaction requires an understanding of the cultural, social, economic and psychological diversity of families, and flexibility to cater to the varied and varying needs of the family, not just the child. By bringing families together to facilitate the sorts of services they need, as well as inter-familial support, FCC aims to build on the strengths of families and children, while addressing their needs, both medical and nonmedical.

Importantly, the family that surrounds the child is not predefined by the practitioner, but is determined by the patient (where they are able to make that determination) and the family themselves (Institute for Patient- and Family-Centred Care, 2010). It may consist of any group of people who have "a continuing legal, genetic, and/or emotional bond" (American Academy of Family Physicians, 2016). As a result, families may or may not include parents, grandparents, siblings, step- or half-siblings, aunts or uncles, or close friends. This broad definition allows the practitioner significant flexibility in who they consider

family for the purposes of delivering care in a family-centred way, but also requires them to put aside preconceptions of who they might consider the family who need to be involved in healthcare decisions. From the perspective of the patient and the family, it allows significant power in who they admit into the clinical relationship, particularly when that determination is reviewed over time, allowing changes in the family to be reflected in the clinical process and family-practitioner relationship.

More recently, the ideas of PCC and FCC have been combined to create the Patient- and Family-Centred Care (PFCC) approach, which, rather than seeing the family itself as the active participant in service delivery, recognises that the patient is at the centre of care, supported and surrounded by their family (Committee On Hospital Care & Institute For Patient- and Family-Centered Care, 2012). By not focusing on the patient or the family exclusively, this model allows patients to drive their own care while still allowing for familial decision-making and support where appropriate. Similar to other developmentally focussed models of family-centred care, this allows practitioners and families to grant the patient increasing levels of control over their own care over time, congruent with their abilities.

By integrating patient-centred and family-centred models together, PFCC can also provide a single approach to care that may be appropriate across the lifespan. As a result, patient- and family-centred models of care have been successfully applied to a range of patients of all ages, with associated increased levels of patient satisfaction (Jakimowicz, Stirling, & Duddle, 2015; Little et al., 2001), rates of mortality (Glickman et al., 2010; Meterko, Wright, Lin, Lowy, & Cleary, 2010), and adherence to treatment (Arbuthnott & Sharpe, 2009; Blackwell, 1996; Garrity, 1981).

A major driver of these improved outcomes may be the inclusion of family members in treatment decisions and the implementation of interventions, which has been shown to be beneficial across aetiologies including diabetes (Hara et al., 2014), breast cancer (Kim &

Morrow, 2007), and substance use disorders (Whitney, Kelly, Myers, & Brown, 2002). Family members can support patients' ability to engage with treatment, can implement interventions outside of the view of the practitioner, and may also bring an intimate perspective on the disease experience to the diagnosis and treatment process that the patient themselves may not have considered.

Significant *third-party disability* has been demonstrated among the families of people living with disability, particularly diseases that impair communication such as hearing loss, aphasia, and dysphagia (Grawburg, Howe, Worrall, & Scarinci, 2013; Nund et al., 2016; Scarinci, Worrall, & Hickson, 2012; Threats, 2010). Third party disability refers to the negative, disabling impacts experienced by a person due to a disease process whose subject is not themselves, such as the hearing loss of a partner, or cerebral palsy experienced by a child (World Health Organisation, 2001). By the close nature of their relationship to the person experiencing the disease process, family members may be more likely to suffer third-party disability. As a result, family members may benefit from supportive interventions in addition to those targeted at their family member's health condition (Threats, 2010).

Patient- and family-centred approaches have been found useful in the management of a variety of chronic diseases (Hudon et al., 2012). However, the chronic nature of these disease processes, in particular their long time courses and the resultant changes in patient health and priorities, can bring significant changes to the nature and implementation of patient-centred approaches in practice (Hudon et al., 2012). The practitioner should be careful to acknowledge the patient's experience not only of their present symptoms but also of the long history of the condition and its management. In addition, practitioners need to be aware of the importance of healthcare transitions, as their patients are unlikely to remain in a single service provision framework for their entire lives, but rather transition between health services many times over the course of their lifespan. A focus on the patient's capability to

manage their own healthcare outside of the practitioner's office can allow a more equal partnership between patients and practitioners, fostering improved engagement with treatment on an ongoing and sustained basis.

Why is hearing loss a chronic disease?

Many hearing losses are chronic diseases: they are incurable in the majority of cases and cause significant ongoing disability across the life course. Like other chronic diseases, they become more common with advancing age, and are likely to coexist with other health conditions, such as cardiovascular disease and diabetes (Abrams, 2017). In fact, hearing loss is one of the most prevalent chronic health conditions in Australia, with more than one in five adult Australians living with a hearing loss, making it more prevalent than asthma, cardiovascular disease, or mental ill health (Access Economics Pty Ltd, 2006, p. 38). Hearing losses caused by genetic mutations or syndromes are a significant cause of hearing-related disability, and are distinct from the more common noise-induced or age-related hearing losses in that they are highly likely to affect children and young adults.

Like many other chronic health conditions, with treatment such as hearing aids, cochlear implants, and other assistive devices, the disability associated with hearing loss may be significantly reduced, leading to improvements in quality of life, social engagement, and other measures of functioning. Among children living with hearing losses, early intervention with hearing aids, cochlear implants, and enriched language input has led to children with hearing losses achieving similar language outcomes to their normally hearing peers (Ching, Dillon, Leigh, & Cupples, 2018). However, even with best practice treatment, significant disability remains, both among older adults (Metselaar et al., 2009; Vuorialho, Karinen, & Sorri, 2006) and among those children whose language outcomes do not "catch up" to those of their peers (Cupples, Ching, Crowe, Day, & Seeto, 2014).

This continuing disability affects the person's general medical care, educational and employment life, and social development and experience. The provision of non-technological rehabilitative treatment can further reduce disability and improve ability to communicate, psychological well-being, and remaining hearing disability (Hickson, Worrall, & Scarinci, 2007).

What evidence is there for PFCC in audiology?

Treatment for chronic hearing losses is long-term, and commonly involves the use of hearing devices. As a part of this, the patient needs to acquire and apply new skills in management and use of these hearing devices. Most hearing aid users have difficulty applying management skills required to effectively manage hearing aids (Bennett, Meyer, Eikelboom, & Atlas, 2018), particularly soon after hearing aid fitting (Solheim, Gay, & Hickson, 2018). Failure to master these skills is a persistent factor in poor adherence to treatment and resultant poor outcomes (Brooks, 1985; Solheim et al., 2018; Sorri, Luotonen, & Laitakari, 1984). People living with hearing loss and hearing aids are also subject to societal stigma (Erler & Garstecki, 2002; Wallhagen, 2010) that can affect their likelihood to seek help for problems related to their hearing loss or hearing aids (Southall, Gagné, & Jennings, 2010), which may be a major factor in their unwillingness to seek help in developing these self-management skills.

Self-management can help patients adhere to treatment, feel more comfortable with the information that they have to learn and apply, and be better able to care for themselves separate from the healthcare provider (Convery, Hickson, Keidser, & Meyer, 2019). By empowering patients to address their own problems outside of the clinic, it can also reduce the time spent between a problem arising and a solution being found. With their focus on personalised delivery, effective and personalised information transfer and ownership and autonomy within the diagnosis and treatment process, patient- and family-centred approaches

to the rehabilitation of chronic hearing losses have the potential to enhance hearing rehabilitation by promoting patient engagement and self-management.

Older adults accessing rehabilitative hearing services in Australia have strong preferences for a patient-centred approach with a strong therapeutic relationship between patient and practitioner at its centre (Grenness, Hickson, Laplante-Lévesque, & Davidson, 2014). This relationship facilitated and was supported by patients receiving increased information about hearing loss, involvement in the treatment decision-making process, and care programmes that were specialised to their personal needs and desires. This model of PCC in audiology is similar to the broader model of PCC put forward by Stewart et al. (2014), which highlights the relationship between patient and practitioner, the development of a shared understanding of the problem and shared determination of management, and engagement with the patient's individual lived experiences as important aspects of PCC.

While Grenness' model was generated by consumers of audiological rehabilitation services, it is clear that this model is, in some ways, aspirational, in that it does not reflect the realities of service being provided to consumers in the clinic. Observations of communication in audiological appointments showed that practitioners did not necessarily allow time for patients to speak, did not engage with their psychosocial needs, did not take opportunities to build interpersonal relationships with their patients, and focused on technological treatment options that were not desired or taken up by a large number of their patients (Grenness, Hickson, Laplante-Lévesque, Meyer, & Davidson, 2015a, 2015b). Grenness et al. (2015a) were careful to stress that audiologists were trying to build relationships with their patients, but that these efforts were stymied by difficulties with interpersonal communication, requiring changes to clinical training and practice to effect appropriately PCC in rehabilitative appointments. Importantly for the present study, it is likely that the same difficulties in delivering patient- and family-centred audiological care to the rehabilitation of

older adults would spread across to other patient populations treated by rehabilitative audiologists in Australia, including emerging adults.

Who are Emerging Adults?

The period between 16 and 25 years of age is one of significant change for many young people. Termed *Emerging Adulthood* by Arnett (2000), it is a stage of life characterised by changes in demographics, a shifting sense of self, and increased development of identity and beliefs. A broad suite of changes in neurology, psychology, and social standing identify the person emerging into adulthood, while still marking them as not yet enjoying the stability that marks full adulthood.

Demographically, many young people in this age group leave home, finish school, embark on post-secondary study, move into the workforce, form long-term romantic relationships, or begin families of their own (National Centre for Vocational Education Research Ltd, 2014). Many of these changes in life circumstance require an increased responsibility for one's own welfare as well as that of others, and so over this period, young adults develop a greater sense of personal awareness and autonomy (Arnett, 2006, p. 13), demanding greater involvement in the making of decisions which affect them. During the transition to emerging adulthood, young people are increasingly self-reliant, rather than depending on parents, teachers, or other support people to provide for their wellbeing, and are more likely to fill these support roles for other people such as their own children or younger siblings.

This increase in responsibility for decisions is supported by neurological development, particularly in the prefrontal cortex, which is responsible for solving complex problems by organising actions in time to achieve specific outcomes (Fuster, 2008, p. 3). This temporal nature of prefrontal cortex function enables planning and assessment of risk not only in the immediate situation, but also over increasing periods, allowing the "playing

forward" of decisions to estimate their impacts on the self and on others into the future. Thus, development of the prefrontal cortex during this period of life increasingly allows emerging adults to integrate information, bringing together varied sources of data to form a rich and detailed picture of the world that they can then use to make decisions going forward (Labouvie-Vief, 2006). This neural development is ongoing through the entire period of emerging adulthood, and so it is vital to recognise not only the benefits to autonomy of emerging adulthood, but also the significant risks to personal safety and wellbeing, both in the short and longer term.

The increased desire by young people for personal autonomy is reflected in increased rates of high-risk behaviours, including high risk sexual behaviour (Dariotis et al., 2008), drug and alcohol use (Furlong, 2009, p. 270), and binge drinking (Australian Bureau of Statistics, 2015). These high-risk behaviours may be indicative of poorer decision-making competence (Goudriaan, Grekin, & Sher, 2007). Poorer decision-making skills and increased impulsivity are associated with poorer adherence to treatment in clinical populations (Barreno et al., 2019), although this has mostly been studied in substance use populations, rather than other chronic health conditions. Supporting patients' ability to exercise autonomy is an important part of PCC (Entwistle & Watt, 2013), and may be particularly important among young adults as their sense of personal autonomy is developing (Arnett, 2000).

The prefrontal cortex is also largely responsible for "theory of mind", or empathy for and cognitive understanding of others (Arnett, 2016). As the prefrontal cortex develops over the period of emerging adulthood, the young person's feelings of attachment to and cognitive understanding of those around them is deepened, tempering the primary impulse to support their own happiness with an increased sense of the wellbeing of those around them (Fuster, 2008, p. 177). This most obviously is reflected in the tendency for emerging adults to engage

in deeper interpersonal relationships, particularly intense and interdependent romantic relationships during this time.

In Australia, more than 8000 young adults, between the ages of 16 and 25, are currently seeking rehabilitative treatment for a hearing loss (Australian Hearing, 2018). While little is known about the aetiologies of these hearing losses, the reducing incidence of curable conductive hearing losses due to otitis media with increasing age (predominately due to the reduction of middle ear pathologies with the maturation of the skull) suggests that many if not most of these hearing losses are either permanent sensorineural or chronic conductive hearing losses, and persistent over time (Epidemiology and Statistics Program, 2012).

What are the challenges for PFCC in Emerging Adults?

The changes inherent in becoming an emerging adult can complicate the implementation of PFCC. Neural development during the period of emerging adulthood leads to the development of higher level thinking, and an increased ability to understand, integrate, and apply complex information, particularly across different areas of life (Fuster, 2008). As a result, over the course of their emerging adulthood, patients develop increased competency to make healthcare decisions that incorporate a range of information and opinion, as well as reflecting their broader priorities and the needs of those around them. This requires practitioners to be aware of the varied informational needs of their patients, both between patients and within a particular patient's healthcare journey over time. Complicating this process, patients' desires for information may not be commensurate with their actual ability to make decisions (Arnett, 2006). Implementing PFCC for emerging adults with hearing loss requires hearing professionals, therefore, to balance patients' desires against their capability to make appropriate decisions about their hearing rehabilitation (Entwistle & Watt, 2013).

In addition, changes in the nature of family may make the incorporation of the family's needs and desires into care problematic (Baas, 2012). For example, at the beginning of emerging adulthood, a young person may consider their significant family members to be their parents and possibly siblings. As they mature, they may move out of home, form a romantic relationship, and perhaps even start a family of their own, and so their romantic partner and children may supplant their parents and siblings as their most significant family members for the purposes of healthcare and decision making (National Centre for Vocational Education Research Ltd, 2014). As a result, practitioners may need to modify their practice to accommodate new parties to the healthcare process.

Why is this thesis needed?

At this stage, little is known about the experiences of young adults living with hearing loss. In particular, it is currently unclear whether hearing losses are having a significant impact on their lives, or what characterises PFCC for them and their families. As a result, it is not known what their preferences might be within a PFCC model, or even whether young people desire a PFCC approach at all. For example, the degree to which family should be involved in care, the levels of information that young people should be given, or whether decision-making should be shared or practitioner-led are all not addressed by the existing literature.

This lack of information puts practitioners at risk of providing care that does not meet the needs of their patients, even while they believe that they are providing a high-quality service. This may compromise care and reduce adherence to and satisfaction with treatment. Disengagement with hearing rehabilitation in young adulthood has the potential to have ongoing consequences for education and employment that are likely to continue into adulthood.

The present study aimed to answer some of these questions so that clinicians working with young adults have an understanding of what domains of care are likely to be important to their patients and their families. By supporting clinicians in this way, not only can there be improvements to the care that is provided to patients, but clinicians can also work in a more effective and fulfilling manner, helping to keep them engaged with the work that they do in providing rehabilitative services. It was envisaged that the research presented in this thesis might help to ensure that services provided to young people are of optimal quality and truly patient-and family-centred.

Thesis

Aims

The aims of this thesis were:

1. To identify the nature of patient- or family-centred care for young adults with chronic diseases.
2. To determine what effect efforts to increase the patient- and family-centredness of care has on the effectiveness of care for young adults with chronic diseases.
3. To explore the functioning of young adult Australians with hearing loss aged 16-25 years as compared to a national sample of young adult Australians in relation to educational achievement, employment engagement and stability, and happiness with aspects of life including overall life satisfaction.
4. To describe the experiences of hearing (re)habilitation for young adults with hearing loss and their utilisation of and satisfaction with hearing devices and services.
5. To determine the association between experiences of audiological (re)habilitation and overall life satisfaction.
6. To explore the perceptions of what constitutes high-quality hearing (re)habilitation according to a group of young adult Australians with hearing loss.

7. To explore the narratives described by mothers of young adults with hearing loss with regards to their involvement with their child's hearing (re)habilitation.

Structure

This study consists of three successive phases, utilising a variety of qualitative and quantitative methods. The use of both qualitative and quantitative methods is useful in health research because it facilitates the expansion of existing knowledge, providing a richer understanding of topics than purely quantitative or qualitative approaches can alone (Padgett, 2012, p. 62).

Phase 1. A systematic review was conducted to achieve Aim 1, and is presented in Chapter 2 of this thesis. As part of this review, the following research question was addressed: "What is the nature of Patient- and Family-Centred Care (PFCC) as defined by young adults living with chronic disease and their family members?" Systematic reviews are an important part of evidence-based practice as they synthesise and critically appraise diverse kinds of literature to develop understandings about the processes underlying clinical practice (Paterson, 2001) in ways that individual studies cannot (Campbell et al., 2011, p. 8). In this case, a qualitative interpretative synthesis methodology was used, adapted from the meta-aggregation methodology proposed by Lockwood, Munn, and Porritt (2015), to collect findings and synthesise them into a coherent structure that can be used by clinicians to better understand, predict, and support their patients' actions (Campbell et al., 2011, p. 105).

Subsequently, a further systematic review was conducted to address Aim 2, discussed in Chapter 3. Utilising a segregated mixed-methods methodology (Sandelowski, Voils, & Barroso, 2006), this review aimed to address the question "What are the impacts of interventions to increase the patient- and family-centredness of care (PFCIs) on the effectiveness of health care provided to young adults living with chronic health conditions?" This review utilised the same qualitative synthesis methodology as the previous, although

with the addition of meta-analysis using random-effects models to combine the quantitative findings (Borenstein, Hedges, Higgins, & Rothstein, 2009).

Phase 2. Based on the preliminary results of the systematic review conducted in Phase 1, an online survey was developed to address Aims 3, 4, and 5. The results of this survey are presented in Chapter 4. Online survey instruments are an appropriate way to collect data from a group of people across a wide geographic range, who are likely to have Internet access, and where identity information is not mandatory for data collection (Sue & Ritter, 2007).

This survey replicated questions posed as part of the *Longitudinal Surveys of Australian Youth* (LSAY, National Centre for Vocational Education Research Ltd, 2014). The LSAY is a large longitudinal survey of a nationally representative sample of 16–25 year-old Australians, and includes information about their educational attainment, employment status, happiness with a variety of aspects of life (including overall life satisfaction), and family structure. Participants were then asked about their experiences of audiological (re)habilitation with a series of questions including the *Measures of Processes of Care for Adults* (MPOC-A, Bamm, Rosenbaum, & Stratford, 2010), which is a measure of the patient-centredness of health care that has been validated in an adult population.

Phase 3. The final phase of this study began with a series of interviews with young adults living with hearing loss to address Aim 6. The interviews were guided by a topic guide that was developed based on the results of Phases 1 and 2. The interviews were conducted and analysed using a Constructivist Grounded Theory methodology (Charmaz, 2006), which is a methodology that aims to understand the mechanisms that underlie social behaviour, in an attempt to understand why they occur and predict them. These interviews were used to construct a model of patient- and family-centred care in this population, which is presented in Chapter 5.

Following these interviews with young adults, a series of interviews were conducted with mothers of these young adult participants, originally in an attempt to address Aim 6 from the perspective of parents. However, when analysing these data, a further aim, reported above as Aim 7, was suggested. Due to the strongly narrative nature of the data elicited from participants, Narrative Inquiry was used to explore the results of these interviews (Clandinin & Connelly, 2000). Narrative Inquiry is a method that focuses on the stories that people construct about their experiences and how these stories exist within their social worlds, and has been used as an effective tool in other health care disciplines, including nursing (Wang & Geale, 2015). The resulting narrative and its implications for clinical practice are discussed in Chapter 6.

Implications, Limitations, and Future Directions. This thesis concludes with a summary of the findings of the empirical work conducted to address the Aims. It also presents a discussion of theoretical and clinical implications of this work, with particular emphasis on implications that are important to PFCC in audiology generally, or for young adults with hearing loss in particular. Finally, limitations of the work are discussed, and some suggestions for future research are presented.

References have been separated by chapter for ease of reading and cross-referencing, and are located at the end of each chapter. Chapters that have been published have minor changes in formatting from the published version to ensure consistency with the Publication Manual of the American Psychological Association (APA) 6th edition throughout (American Psychological Association, 2009). As each of the chapters 2–6 were prepared with the intention that they be submitted to a peer-reviewed journal, there is some duplication in background and context for each paper.

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CHAPTER 2 - THE NATURE OF PATIENT- AND FAMILY-CENTRED CARE FOR YOUNG ADULTS LIVING WITH CHRONIC DISEASE AND THEIR FAMILY MEMBERS: A SYSTEMATIC REVIEW (PUBLISHED)

Rationale

As hearing loss is a chronic health condition, and as the "chronicness" of health conditions has been established to have an impact on the nature of PFCC, it was important to consider the literature about PFCC in other populations of young people living with chronic health conditions. The literature was varied, and of highly variable quality, and no previous systematic review to address this question could be found. As a result, a systematic review of the literature was conducted and has been published in the International Journal of Integrated Care (D. Allen, Scarinci, & Hickson, 2018). This publication is presented in full in this chapter, with the only modifications being that it has formatted to be consistent with the APA Publication Manual, 6th Edition (American Psychological Association, 2009).

Due to the restrictions of the journal submission process, it was not possible to present an in-depth discussion of the process of critical appraisal as it was undertaken as part of this systematic review. However, it should be noted that the use of critical appraisal tools in systematic reviews incorporating qualitative literature is controversial (Barbour, 2001; Popay, Rogers, & Williams, 1998) largely due to the diversity of epistemological and practical approaches (Caracelli & Cooksy, 2013; Eakin & Mykhalovskiy, 2003). However, higher-quality qualitative studies are more useful for interpretative synthesis (Carroll, Booth, & Lloyd-Jones, 2012), and so the use of critical appraisal tools to identify and filter out poor-quality reports of qualitative studies is recommended by the Cochrane Collaboration (Hannes, 2011). Due to the large amount of data identified as part of the present review, and the subjective variation in quality of the reports, it was decided to use a set of critical appraisal

tools as a screening tool to identify high-quality reports. Initially, three sets of quality appraisal tools were identified as potentially applicable – the Critical Appraisal Skills Program (CASP) checklists (2013a, 2013b, 2013c, 2013d, 2013e), the Qualitative Appraisal and Review Instrument (QARI) published by the Joanna Briggs Institute (2014), and the University of Salford tool for the analysis of qualitative studies (Long, Godfrey, Randall, Brettle, & Grant, 2002). Of these, the CASP checklists were chosen as they have been used in a variety of existing systematic reviews, and do not rely on extensive theoretical knowledge of qualitative research (Hannes, Lockwood, & Pearson, 2010). The ways in which the CASP tools were modified and used for critical appraisal are detailed in the chapter.

While this systematic review broadened its focus to a range of chronic health conditions, its results also fit well with models of patient-centred care developed among those living with hearing loss. Grenness, Hickson, Laplante-Levesque, and Davidson (2014) studied a group of older adults living with hearing loss, and highlighted the importance of addressing the unique and changeable needs of the patient and their family and the importance of the therapeutic relationship, as well as the importance of practitioner professionalism and having more than merely technical skill. Grenness et al. also highlighted the provision of relevant information in an honest and complete manner as an important element of patient control and efficacy, similar to the present results. However, as might be expected due to the older age of their participants, Grenness et al.'s respondents did not experience the same kinds of tensions with family members that were reported in the present study's literature relating to young adults, and so this is not reflected in their model.

Introduction

Active integration of patients into the process of health care delivery is one of the central tenets of patient-centred care (PCC; M. Stewart et al., 2014), a model of health care that prioritises patients' desires and individual needs (Laine, 1996). While various models

and definitions of PCC exist, the common characteristics of all these models and definitions is patient empowerment through the provision of information and opportunities to patients. This enables them to actively participate in choosing and guiding their care, supported by strong relationships with their practitioners (Mead & Bower, 2000).

The benefits of patient-centred approaches are particularly evident in chronic disease management, as care must be provided over an extended period, and so small changes in patient engagement can have wide-ranging effects on the quality of care long-term. Positive effects have been reported across a variety of aetiologies, with reductions in morbidity or costs of care demonstrated in patients with migraine (Diamond, Wenzel, & Nissan, 2006), diabetes and hypertension (Or & Tao, 2016), eating disorders (Anastasiadou, Medina-Pradas, Sepulveda, & Treasure, 2014), infertility (Gameiro, Canavarro, & Boivin, 2013), and heart failure (Sahlen, Boman, & Brannstrom, 2016).

When treating younger people and children, the patient-centred approach is often extended to include the family in decision-making and treatment delivery, an approach known as family-centred care (Rosenbaum, King, Law, King, & Evans, 2009). Family-centred approaches hold that families, like patients themselves, are unique, with different strengths and weaknesses, and that the most effective way to treat children's illnesses is through engaging with and treating the family as a unit (R. I. Allen & Petr, 1996). The role of the practitioner in family-centred care is to build a relationship with the family so that the practitioner and the family work together towards beneficial outcomes for the patient (Epley, Summers, & Turnbull, 2010).

Over recent years, the term "patient- and family-centred care" (PFCC) has been put forward by the American Academy of Paediatrics, to reflect the changing capabilities of young patients as they grow older (Committee On Hospital Care & Institute For Patient- and Family-Centered Care, 2012). This combined term emphasises the patient as being able to

participate in their care, with the support of their families. It blends both patient-centred and family-centred approaches together to treat both the patient and their family members in the health care process. PFCC can be a powerful way of improving communication between patient and the health care team, breaking down potential barriers between the two and making the patient a more equal and active participant in the health care team (Mead & Bower, 2000). This improved communication facilitates the free exchange of information between patient and practitioner so that both can express treatment preferences and together come to a decision about treatment progress; a process called shared decision-making (Charles, Whelan, & Gafni, 1999). Shared decision-making leads to higher quality decisions, improved adherence to treatment, and more satisfactory outcomes (Desroches, 2010). Shared decision-making is particularly important for those living with chronic diseases, defined as diseases that persist over a long time course, that are recurrent, and where the focus is on symptom management rather than curing the underlying disease process (O'Halloran, Miller, & Britt, 2004).

The dominant models of patient-centred care have traditionally been developed in family medicine (M. Stewart et al., 2014) and general practice (Mead & Bower, 2000). As a result, these models have focused on the patient populations who attend family and general practices, predominately adults over the age of 25 (Britt et al., 2015). Similarly, the development of models of family-centred care has primarily been in the context of younger children attending health services with their parents (Rosenbaum et al., 2009). Patients between adolescence and adulthood have not been regularly engaged in research determining the nature of PFCC.

Arnett (2006, pp. 5-6) describes the transition from adolescence to young adulthood, which he terms "emerging adulthood", as a period traditionally marked by changes in demographics, self-identity, and ideology (Arnett, 2000). Demographically, the beginning of

emerging adulthood is a time of transition, where young adults may transition to tertiary or vocational study, begin their working lives, and move out of the home, and away from their caregivers - potentially to form romantic relationships or families of their own (National Centre for Vocational Education Research Ltd, 2014). Regarding self-identity, emerging adulthood is marked specifically by a development of the sense of self as an individual, separate from family. As part of this process, emerging adults claim from their parents and caregivers the rights to hold their health care information, and to use it to make decisions about their lives and bodies (Koepke & Denissen, 2012). These sorts of changes, particularly changes in the composition of the family unit, have important implications for the design and implementation of PFCC.

Many emerging adults are living day-to-day while managing a variety of chronic health conditions, for example cardiovascular disease, chronic kidney disease, diabetes (Australian Institute of Health and Welfare, 2014), HIV/AIDS (Centres for Disease Control and Prevention, 2008; Murray et al., 2012; UNAIDS, 2015), congenital disorders such as haemophilia (Soucie, Evatt, & Jackson, 1998), and mental and behavioural disorders including eating disorders (Hudson, Hiripi, Pope, & Kessler, 2007), depression, and anxiety (Kessler, 1994; Kessler et al., 2005).

It has been shown that PFCC in chronic disease care requires practitioners to take a role in legitimising and validating the patient's experience of illness, encouraging hope for the future, and advocating for their rights on an ongoing basis (Hudon et al., 2012). As these aspects of PFCC are driven by the continuing nature of the chronic condition and the requirement for the patient to manage their health on an ongoing basis, they are not universally present in acute care. As such, investigation in chronic care specifically is necessary to bring forward these factors.

It is currently unclear what young adults living with chronic diseases and their family members identify as PFCC, and the research on this topic is diverse. Systematic review and synthesis can enhance the usefulness of reports of diverse kinds of research in clinical practice (Sandelowski & Barroso, 2007, pp. 9-10), by developing theories about the processes underlying these various studies (Paterson, 2001, p. 14) in a way that would not be possible for any single study alone (Campbell et al., 2011, p. 8). By focusing on emerging adults living with chronic disease, this review aims to determine an understanding of PFCC that addresses the needs of them and their families.

The aim of this systematic review is to interpretatively synthesise reports of studies addressing the nature of PFCC among emerging adults living with chronic disease and their family members to generate a conceptual understanding of the topic. The research question for the review was "What is the nature of PFCC as defined by young adults living with chronic disease and their family members?"

Method

Research Team

This review was conducted by the three authors. The first and third authors are audiologists, and the second a speech-language therapist, all with clinical and research experience. This work forms part of the PhD studies of the first author.

Data Sources

A range of search strategies were used to identify a broad range of literature for consideration for this review. The search strategies for this systematic review are presented in Table 2.1. CINAHL Terms and MeSH Headings relevant to PFCC, emerging adults, and adolescents were identified, and these were used for initial searches in CINAHL Complete and MEDLINE. From this, additional keywords were identified, which were incorporated

into search strategies for MEDLINE (via EBSCOHost), CINAHL Complete (via EBSCOHost), PsycINFO and EMBASE in consultation with a librarian specialising in health sciences. Due to the discussion of PCC over a long period of time in the psychological literature, no date restriction was placed on the searches.

Table 2.1

Search strategies used to identify papers.

Database	Search Date	Search Term	Total	Retained
MEDLINE	15/09/15	(MH Adolescent OR MH Young Adult OR MH Transition to Adult Care) NOT (MH Aged OR MH Middle Aged) AND (MH Patient-Centered Care OR MH Professional-Family Relations OR MH Personal Autonomy OR MH Patient Participation OR MH Professional-Patient Relations)	4802	187
EMBASE	21/09/15	('adolescent'/exp OR 'young adult'/exp OR 'transition to adult care'/exp) NOT ('aged'/exp OR 'middle aged'/exp) AND ('holistic care'/exp OR 'patient decision making'/exp OR 'patient autonomy'/exp OR 'personal autonomy'/exp OR 'family centered care'/exp OR 'patient participation'/exp OR 'doctor patient relation'/exp) AND [embase]/lim	1853	213
CINAHL	22/09/15	((MH "Adolescence") OR (MH "Young Adult")) NOT ((MH "Aged") OR (MH "Middle Age")) AND ((MH "Professional-Patient Relations") OR (MH "Physician-Patient Relations") OR (MH "Patient Centered Care") OR (MH "Professional-Family Relations") OR (MH "Family Centered Care") OR (MH "Patient Autonomy") OR (MH "Decision Making, Patient"))	3025	210

Database	Search Date	Search Term	Total	Retained
PsycINFO	28/09/15	Index Terms: "client centered therapy" OR Index Terms: "client participation" OR Index Terms: "self determination" OR FirstPage: "patient-centered" OR FirstPage: "patient-centred" OR FirstPage: "patient centered" OR FirstPage: "patient centred" OR FirstPage: "person-centered" OR FirstPage: "person centered" OR FirstPage: "person centred" OR FirstPage: "person-centred" OR FirstPage: "family-centred" OR FirstPage: "family centred" OR FirstPage: "family-centered" OR FirstPage: "family centered" OR FirstPage: "physician-patient" OR FirstPage: "physician-family" OR FirstPage: "practitioner-patient" OR FirstPage: "practitioner-family" OR FirstPage: "clinician-patient" OR FirstPage: "clinician-family" OR FirstPage: "shared decision making" AND Age Group: Adolescence (13 to 17 yrs) OR Young Adulthood (18 to 29 yrs) AND NOT Age Group: Neonatal (birth to 1 mo) OR Infancy (2 to 23 mo) OR Preschool Age (2 to 5 yrs) OR Middle Age (40 to 64 yrs) OR Aged (60 yrs & older) OR Very Old (85 yrs & older)	935	56
Articles identified by hand			50	50
Total			10665	716

Study Selection

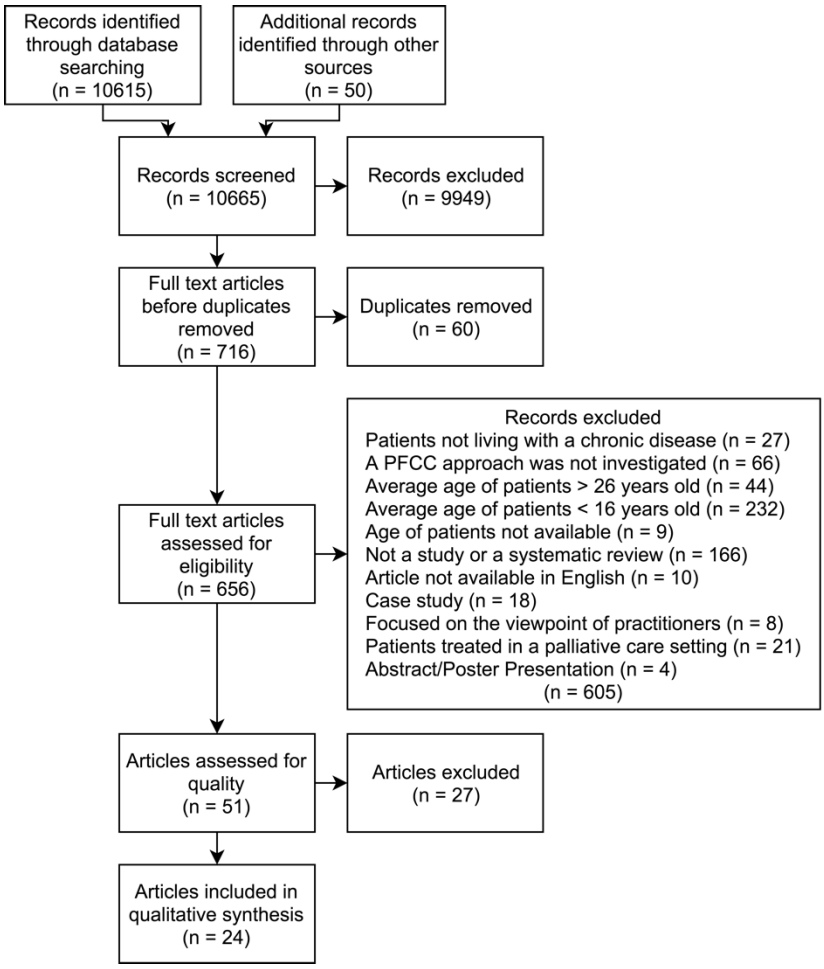

Figure 2.1. Flow of articles through appraisal process

The movement of reports through the search, selection, and quality appraisal processes can be seen in Figure 2.1. From initial searches, 10,615 reports were identified. Fifty reports were also identified through the researcher's existing library and by scanning reference lists. These papers were then screened to identify papers which could be relevant to the research question, based on the title and abstract, with 716 remaining. Sixty-six duplicates were removed. The text of each of these was obtained, and a further screening process was undertaken based on the following inclusion criteria:

a) Patients were living with a chronic disease

b) The nature of a PFCC approach was investigated

c) Average age of patients < 26 years old

d) Average age of patients > 16 years old

e) The perspective of either patients or their family members were investigated, or a systematic review of such studies was conducted

f) Article was available in English

g) At least three participants were involved

h) The study did not focus exclusively on the viewpoint of practitioners

i) Patients were not being treated in a palliative care setting

A total of 51 papers passed this screening process and proceeded to the quality appraisal stage, including 46 reports of qualitative studies, two reports of quantitative studies, and three systematic reviews of published literature. In order to ensure that the inclusion criteria were applied appropriately, of the 656 papers identified for full-text checking, 10% (n = 66) were randomly selected, and independently assessed against the eligibility criteria by the second and third authors for agreement on their inclusion or exclusion. There was disagreement on three papers, which were then discussed and consensus was reached.

Quality Appraisal

The use of critical appraisal tools in systematic reviews incorporating qualitative literature is controversial (Barbour, 2001; Popay et al., 1998) largely due to the diversity of epistemological and practical approaches (Caracelli & Cooksy, 2013; Eakin & Mykhalovskiy, 2003). However, higher-quality qualitative studies are more useful for interpretative synthesis (Carroll et al., 2012), and so the use of critical appraisal tools to identify and filter out poor-quality reports of qualitative studies is recommended by the Cochrane Collaboration (Hannes, 2011). Due to the large amount of data identified as part of the present review, and the subjective variation in quality of the reports, it was decided to use a set of critical appraisal tools as a screening tool to identify high-quality reports.

Initially, three sets of quality appraisal tools were identified as potentially applicable – the Critical Appraisal Skills Program (CASP) checklists (2013a, 2013b, 2013c, 2013d, 2013e), the Qualitative Appraisal and Review Instrument (QARI) published by the Joanna Briggs Institute (2014), and the University of Salford tool for the analysis of qualitative studies (Long et al., 2002). Of these, the CASP checklists were chosen as they have been used in a variety of existing systematic reviews, and do not rely on extensive theoretical knowledge of qualitative research (Hannes et al., 2010). After a review of systematic reviews using the CASP checklists, it was decided that papers would have to score a "Yes" on every relevant question of the appropriate CASP checklist to proceed to data extraction. After piloting the Randomised Controlled Trial (Critical Appraisal Skills Program, 2013c), Case Control (Critical Appraisal Skills Program, 2013d), and Cohort (Critical Appraisal Skills Program, 2013e) CASP checklists, the Randomised Controlled Trial checklist was found to be too restrictive, and so the Cohort and Case Control checklists were combined, and an ethics criterion added, to form a Quantitative Research Study checklist. The modified tools are listed in Table 2.2.

Table 2.2

Modified CASP Tools used for quality appraisal

Qualitative Research Study	Qualitative Systematic Review
1. Was there a clear statement of the aims of the research? 2. Is a qualitative methodology appropriate? 3. Was the research design appropriate to address the aims of the research? 4. Was the recruitment strategy appropriate to the aims of the research? 5. Was the data collected in a way that addressed the research issue? 6. Has the relationship between researcher and participants been adequately considered? 7. Have ethical issues been taken into consideration? 8. Was the data analysis sufficiently rigorous? 9. Is there a clear statement of findings?	1. Did the review address a clearly focused question? 2. Did the authors look for the right type of papers? 3. Do you think all the important, relevant studies were included? 4. Did the review's authors do enough to assess the quality of the included studies? 5. If the results of the review have been combined, was it reasonable to do so?

Quantitative Research Study
1. Did the study address a clearly focused issue? 2. Did the authors use an appropriate method to answer their question? 3. Were the cases recruited in an acceptable way? 4. Were the controls selected in an acceptable way? 5. Was the exposure accurately measured to minimise bias? 6. Was the outcome accurately measured to minimise bias? 7. Have the authors taken account of the potential confounding factors in the design or in their analysis? 8. Was the follow up of subjects complete enough? Was the follow up on subjects long enough? 9. Have ethical issues been taken into consideration? 10. Do you believe the results?

The results of the application of the CASP tools for the 51 papers that passed to the quality appraisal stage can be seen in Table 2.3, Table 2.4, and Table 2.5. In each table, "Y" indicates a positive answer to the relevant question, "N" indicates a negative answer, and "?"

indicates that it was unclear whether the response should be positive or negative. Further clarification of unclear responses was not required, as each of these papers had already been excluded by a clear negative answer elsewhere in the tool.

Of three systematic reviews identified in the search, two were excluded as neither had usilised a quality appraisal methodology. Of 46 qualitative studies, 23 were excluded. The most common reason for exclusion was insufficient consideration of the relationship between researcher and participants (n = 13), either by not explicating the position of the researcher in the research or by not taking steps to reduce the researcher's effect on the results, such as independent coding of data by more than one researcher or review of coding by others on the research team. Several papers were also excluded due to a failure to report findings using the voices of the participants (n = 6), or for not collecting the data in an appropriate fashion (n = 5). The two quantitative reports were both excluded after quality appraisal due to insufficient consideration of potential confounding factors. The 24 remaining papers, which were all reports of qualitative papers or systematic reviews of qualitative papers, proceeded to synthesis.

Of the 51 papers that proceeded to quality appraisal, 12 (24%) were randomly selected, and these were independently assessed by the second and third authors according to the quality appraisal tools. There was disagreement on two papers, which were then discussed until consensus was reached.

Data Extraction and Synthesis

Initially, each of the 24 full papers was read by the first author, and general details of each paper were recorded, including the number of participants, their relationship to the patients, the patients' diagnoses, the data collection method and analysis style, and a broad outline of the findings. These may be seen in Table 2.6.

As all 24 papers that passed the quality appraisal step were qualitative papers or a systematic review of qualitative papers, a meta-aggregation methodology, adapted from that proposed by Lockwood et al. (2015), was chosen due to its applicability to a variety of types of papers. In this method, each paper was read, and the findings identified and extracted, along with a unit of data (in this case, a quote from a participant) that supported each finding. Findings that were unsupported by data were not recorded, and where practitioners were also involved as participants in a study, only statements attributable to patients or family members were used to identify findings and associated data. These findings were then collated into groups of similar findings, from which overall themes were synthesised. In several cases, no verbatim quote from the paper could be found that adequately summarised the finding, and so Lockwood's methodology was modified by allowing the researchers to reword findings slightly to reflect the context of the report in which the finding was identified.

Table 2.3

Quality appraisal results for assessed systematic reviews

Systematic Review	**Q1**	**Q2**	**Q3**	**Q4**	**Q5**	**Included**
Anastasiadou et al. (2014)	Y	Y	Y	N	N	N
Fegran, Hall, Uhrenfeldt, Aagaard, and Ludvigsen (2014)	Y	Y	Y	Y	Y	Y
Hussen et al. (2014)	N	Y	?	N	Y	N

"Y" = Yes, "N" = No, "?" = question was unable to be answered clearly in this case

Table 2.4

Quality appraisal results for assessed quantitative papers

Quantitative Report	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Included
Mauerhofer, Bertchold, Akré, Michaud, and Suris (2010)	Y	Y	Y	Y	N	N	N	Y	Y	Y	N
Sonneveld, Strating, van Staa, and Nieboer (2013)	Y	Y	Y	N	Y	Y	N	N	Y	Y	N

"Y" = Yes, "N" = No, "?" = question was unable to be answered clearly in this case

Table 2.5

Quality appraisal results for assessed qualitative papers

Qualitative Report	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Included
Brumfield and Lansbury (2004)	Y	Y	Y	Y	Y	N	Y	Y	Y	N
Cochrane et al. (2015)	Y	Y	Y	?	?	N	?	?	Y	N
Darrah, Magil-Evans, and Adkins (2002)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Davis-Brown, Carter, and Miller (2012)	Y	Y	N	N	N	N	N	N	N	N
Delman, Clark, Eisen, and Parker (2015)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Dogba et al. (2014)	Y	Y	Y	Y	Y	Y	Y	?	N	N
Doig, Fleming, Cornwell, and Kuipers (2009)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Dovey-Pearce, Hurrell, May, Walker, and Doherty (2005)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Dunsmore and Quine (1995)	Y	Y	Y	Y	Y	Y	N	Y	N	N
Dupuis, Duhamel, and Gendron (2011)	Y	Y	Y	Y	Y	N	Y	Y	Y	N
Fair, Sullivan, Dizney, and Stackpole (2012)	Y	Y	Y	Y	Y	N	Y	?	Y	N
Garvie et al. (2009)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Gerten and Hensley (2014)	N	Y	N	N	N	Y	Y	Y	Y	N
Gillard and Roark (2013)	Y	Y	Y	Y	N	Y	Y	N	N	N
Gilmer et al. (2012)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Grealish, Tai, Hunter, and Morrison (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Qualitative Report	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Included
Harper, Dickson, and Bramwell (2014)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Hauser and Dorn (1999)	Y	Y	Y	Y	Y	?	Y	N	Y	N
Honey et al. (2008)	Y	Y	N	Y	N	?	?	Y	Y	N
Larivière-Bastien, Bell, Majnemer, Shevell, and Racine (2013)	N	Y	N	N	Y	N	Y	Y	Y	N
Ledford (2015)	Y	Y	Y	N	Y	N	Y	Y	Y	N
Lee et al. (2006)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Lester et al. (2011)	Y	Y	Y	Y	Y	N	Y	Y	Y	N
Lewis and Noyes (2013)	Y	Y	N	Y	N	Y	Y	Y	Y	N
Lucksted et al. (2015)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Miles, Edwards, and Clapson (2004)	Y	Y	Y	Y	Y	N	Y	N	Y	N
Munson et al. (2012)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Nilson et al. (2012)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Offord, Turner, and Cooper (2006)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Olsen and Sutton (1998)	N	Y	N	N	Y	N	Y	Y	Y	N
Parron (2014)	Y	Y	?	?	?	?	?	N	Y	N
Patterson and Lanier (1999)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Price et al. (2011)	Y	Y	Y	Y	Y	N	Y	Y	Y	N
Racine, Lariviere-Bastien, Bell, Majnemer, and Shevell (2013)	Y	Y	Y	N	Y	N	N	Y	Y	N
Reiss, Gibson, and Walker (2005)	Y	Y	Y	Y	Y	Y	Y	Y	N	N
Rudgley (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Rydström, Ygge, Tingberg, Navèr, and Eriksson (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Saaltink, Mackinnon, Owen, and Tardif-Williams (2012)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sasse, Aroni, Sawyer, and Duncan (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sawin et al. (2015)	N	Y	N	N	Y	N	Y	Y	Y	N
Shaw, Southwood, and McDonagh (2004)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sly et al. (2014)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
D. A. Stewart, Law, Rosenbaum, and Willms (2001)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Swift et al. (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Qualitative Report	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Included
van Staa, Jedeloo, van Meeteren, and Latour (2011)	Y	Y	Y	Y	Y	Y	Y	Y	N	N
Webster and Harrison (2008)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

"Y" = Yes, "N" = No, "?" = question was unable to be answered clearly in this case

Table 2.6

General details of included papers

Paper	Aetiology	Included Groups	Participants	Patient age - mean [range] (sd)	Phenomenon of interest	Method of Data Collection	Method of Data Analysis	Primary Findings
Darrah et al. (2002)	Cerebral Palsy	Emerging Adults, Parents	38 families	[19–23]	Satisfaction with care	Questionnaire, Interview	Content Analysis	Caring and Supportive People Fighting and Fatigue Communication and Information Disability Awareness
Delman et al. (2015)	Serious Mental Illness	Emerging Adults	24 patients	24 [19–30]	Facilitators and Barriers to shared decision-making	Interview	Inductive Thematic Analysis	Facilitators: • psychiatrist's interest in the patient's perspective • support of other mental health providers • personal growth • self-confidence • greater availability of the psychiatrist Barriers: • short duration of meetings • Psychiatrist's resistance to the patient's perspective • limited self-efficacy
Doig et al. (2009)	Traumatic Brain Injury	Emerging Adults, Parents	12 patients and parents. Three therapists also interviewed.	24.7 (6.9)	Experience of a goal-directed therapy programme	Interview	Framework method	Provision of Structure Goals and Motivation Goal ownership Impact of awareness on participation Challenges Family Involvement Satisfaction and Progress Cognitive Function Goal Evolution Priorities

Paper	Aetiology	Included Groups	Participants	Patient age - mean [range] (sd)	Phenomenon of interest	Method of Data Collection	Method of Data Analysis	Primary Findings
Dovey-Pearce et al. (2005)	Diabetes	Emerging Adults	19 patients (interviews) 8 patients (focus groups)	19.9 (3.12) for interviews; 19.4 (2.67) for focus groups	Suggestions for appropriate diabetes service	Interview; Focus Group	Framework Approach	Diagnosis Continuity of staff contact Influence of age on care Interactions with staff Access and Environment Suggestions for service development
Fegran et al. (2014)	Various	Emerging Adults	18 studies - metasynthesis		Adolescents and Young Adults transition experiences	Literature search	Qualitative Metasynthesis	Facing changes in significant relationships Moving from a familiar to an unknown ward culture Being prepared for transfer Achieving responsibility
Garvie et al. (2009)	HIV-1	Emerging Adults	17 patients	19.93 (1.29) [17.6–22.5]	Suggestions for appropriate modified directly observed therapy (MDOT) adherence intervention.	Focus Group	Content Analysis	Barriers to adherence MDOT Provider characteristics Location and safety of MDOT interactions Communication between MDOT provider and participant Logistics of MDOT interactions Duration of MDOT intervention Additional services to be provided during MDOT interaction Feasibility and acceptance of MDOT program Potential barriers to MDOT program
Gilmer et al. (2012)	Mental Health Disorders	Emerging Adults, Parents	75 patients, 14 parents	[18–24]	Needs for Mental Health and other services	Focus Group	Inductive Thematic Analysis	Mental health and substance abuse services Services that foster a transition to independence
Grealish et al. (2013)	Psychosis	Emerging Adults	9 patients	16.4	Empowerment from the perspective of young people with psychosis	Interview	Interpretative Phenomenological Analysis	Individual control and choice vs inflexibility Being listened to, respected, and validated Communication Response of services Coping and structure Quality of relationship and support

Paper	Aetiology	Included Groups	Participants	Patient age - mean [range] (sd)	Phenomenon of interest	Method of Data Collection	Method of Data Analysis	Primary Findings
Harper et al. (2014)	Mental Health Disorders	Emerging Adults	10 patients	[16–18]	Experiences of 16–18 Mental Health Service	Interview	Interpretative Phenomenological Analysis	Developmentally attuned services Power differentials Parental involvement Developing self-expression Continuity and loss of relationships
Lee et al. (2006)	Mental Health Disorders	Emerging Adults	389 patients	17	Attitudes towards mental health services among young adults in foster care	Interview	Thematic Analysis	Positive experiences are associated with beneficial care and relationships with a mental health professional; negative experiences were associated with concerns about treatment, poor relationships with a mental health professional, and unprofessional practice.
Lucksted et al. (2015)	Psychosis	Emerging Adults	32 patients	23 [<20–34]	Views of engagement in an early intervention program for psychosis	Interview	Thematic Analysis	Individualised care • Focus on life goals • Effectiveness • Warm respect Program attributes • Team structure • Setting and location • Medication management approach • Active outreach Family member influences • Promoting engagement • Deterring engagement • Personal attributes
Munson et al. (2012)	Mood Disorders	Emerging Adults	60 patients	20.97 (2.08)	Experiences of mental health service use during the transition to adulthood.	Interview	Grounded Theoretic Analysis	Dynamic nature of service utilisation over time Core factors that impact service use at any given time
Nilson et al. (2012)	Haemophilia	Emerging Adults	18 patients	25.2 [17–31]	Health care-related knowledge, attitudes, and behaviours of young men with haemophilia	Interview (face to face and by telephone)	Constant Comparative Method	Reluctance to acknowledge having mild haemophilia Experiential learning trumps advice from the haemophilia team Negative reception to the health care teams' approaches Strategies for managing potential bleeds: watch and wait

Paper	Aetiology	Included Groups	Participants	Patient age - mean [range] (sd)	Phenomenon of interest	Method of Data Collection	Method of Data Analysis	Primary Findings
Offord et al. (2006)	Anorexia Nervosa	Emerging adults	7 patients	[16–23]	Experiences of treatment and discharge of young adults in inpatient treatment for anorexia nervosa	Interview	Interpretative Phenomenological Analysis	Removal from Normality vs. connecting with the outside world • Suspension of real life • Normality around mealtimes • Suspension in development, compounding a sense of isolation • Contrasts in structure and support Treated as another anorexic vs. a unique individual in distress • Staff assumptions about eating disorders • Standardised programmes • Physical recovery prioritised over psychological recovery • Recognising the eating disorder as a symptom • A genuinely holistic approach Control and collaboration • Initial taking away of control • A structured containment • Powerlessness, punishment and inadequacy • Doing battle • Collaborating in one's own care • Collaborating within therapy • Preparing for discharge - handing back control The importance of peer relationships • Distance from peers in the outside world • Being alongside peers in distress - acceptance versus segregation • Being alongside peers with anorexia nervosa - identification versus competition
Patterson and Lanier (1999)	Special Health Care Needs (Chronic illnesses or physical disabilities)	Emerging Adults	7 patients	24.3 (6.47) [17–33]	Experiences of, and facilitators and barriers to transition from paediatric care to adult care.	Focus Group	Grounded Theoretic Analysis	Barriers to successful transition Burned out on health care What helps or might have helped transition Strategies for successful transition

Paper	Aetiology	Included Groups	Participants	Patient age - mean [range] (sd)	Phenomenon of interest	Method of Data Collection	Method of Data Analysis	Primary Findings
Rudgley (2013)	Attention Deficit Hyperactivity Disorder	Emerging Adults	4 patients	18.5, 19, 19, 19	Experiences of transition from paediatric to adult care of young adults with ADHD	Interview	Interpretative Phenomenological Analysis	Personal experience of ADHD diagnosis and treatment Impact of ADHD on self and relationships Living with ADHD Moving on
Rydström et al. (2013)	HIV	Emerging Adults	10 patients	18 [15–21]	Experiences of young people growing up with innate or early acquired HIV infection	Interview	Content Analysis	To protect oneself from the risk of being stigmatised To be in control Losses in life, but HIV is not a big deal Health care/health care providers Belief in the future
Saaltink et al. (2012)	Intellectual Disabilities	Emerging Adults, Parents, Siblings	4 patients, 4 mothers, two siblings	[14–18]	The negotiation of the right to participate in shared decision making in a family context.	Interview	Interpretative Phenomenological Analysis	Autonomous participation Participation and protection: guidance and parents' choice Decision-making processes as normal and natural Enabled choices
Sasse et al. (2013)	Various chronic issues, particularly eating disorders	Parents	17 parents	16 (1.4) [13–18]	Parental perspectives on confidential consultations between their adolescent children and health care providers	Interview	Content and Thematic Analysis	Variation in parental views about confidential consultations for adolescents The role of a parent: • essential to their child • expert on their child • legal guardian of their child The influence of trust
Shaw et al. (2004)	Juvenile Idiopathic Arthritis	Emerging Adults, Parents	12 adolescents, 14 parents of adolescents, 18 young adults, 9 parents of young adults.	16 [13–18] (adolescents); 23 [19–30] (young adults);	Experiences of transitional care for adolescents with juvenile idiopathic arthritis.	Focus Groups	Interpretative Phenomenological Analysis	Transitional care: • multi-dimensional • coordinated • supportive • developmentally appropriate • age-appropriate Transfer from paediatric to adult services Preparation for transfer
Sly et al. (2014)	Anorexia Nervosa	Emerging Adults	8 patients	25 [18–34]	Experiences of therapeutic alliance during in-patient treatment for anorexia nervosa	Interview	Interpretative Phenomenological Analysis	Alliance as a key experience Active, not passive Taboo talking First impressions count

Paper	Aetiology	Included Groups	Participants	Patient age - mean [range] (sd)	Phenomenon of interest	Method of Data Collection	Method of Data Analysis	Primary Findings
D. A. Stewart et al. (2001)	Physical disabilities	Emerging Adults, Parents	21 patients; 12 parents. One service provider also interviewed	23.2 [19–30]	Experiences of transition for young people with physical disabilities.	Interview	Editing style of thematic analysis	The Context: “Trying to Fit” The Transition Process: “Changes and Cliffs” Needs and Services: “Building a Bridge”
Swift et al. (2013)	Attention Deficit Hyperactivity Disorder	Emerging Adults	10 patients	[17–18.5]	Experieces of transition to adult mental health services	Interview	Thematic Analysis	Clinician qualities and relationship Responsibility of care Nature and severity of problems Expectations of AMHS.
Webster and Harrison (2008)	Mental Health Disorders	Emerging Adults	20 patients	[18–25]	Experiences of the onset of mental health problems, and initial interactions with the health system	Interview	Grounded Theoretic Analysis	First sign Recognition Understanding Resolution Maze to care model

Results

The results of the qualitative synthesis indicated that emerging adult patients with chronic diseases and their families experienced high-quality PFCC as having three major characteristics: (1) patients and practitioners felt able to engage with each other on an emotional and social level; (2) patients and families felt empowered to be part of the care process; and (3) patients and families experienced care as effective. These themes and subthemes are depicted in Figure 2.2. Each characteristic, with relevant subthemes, is discussed below.

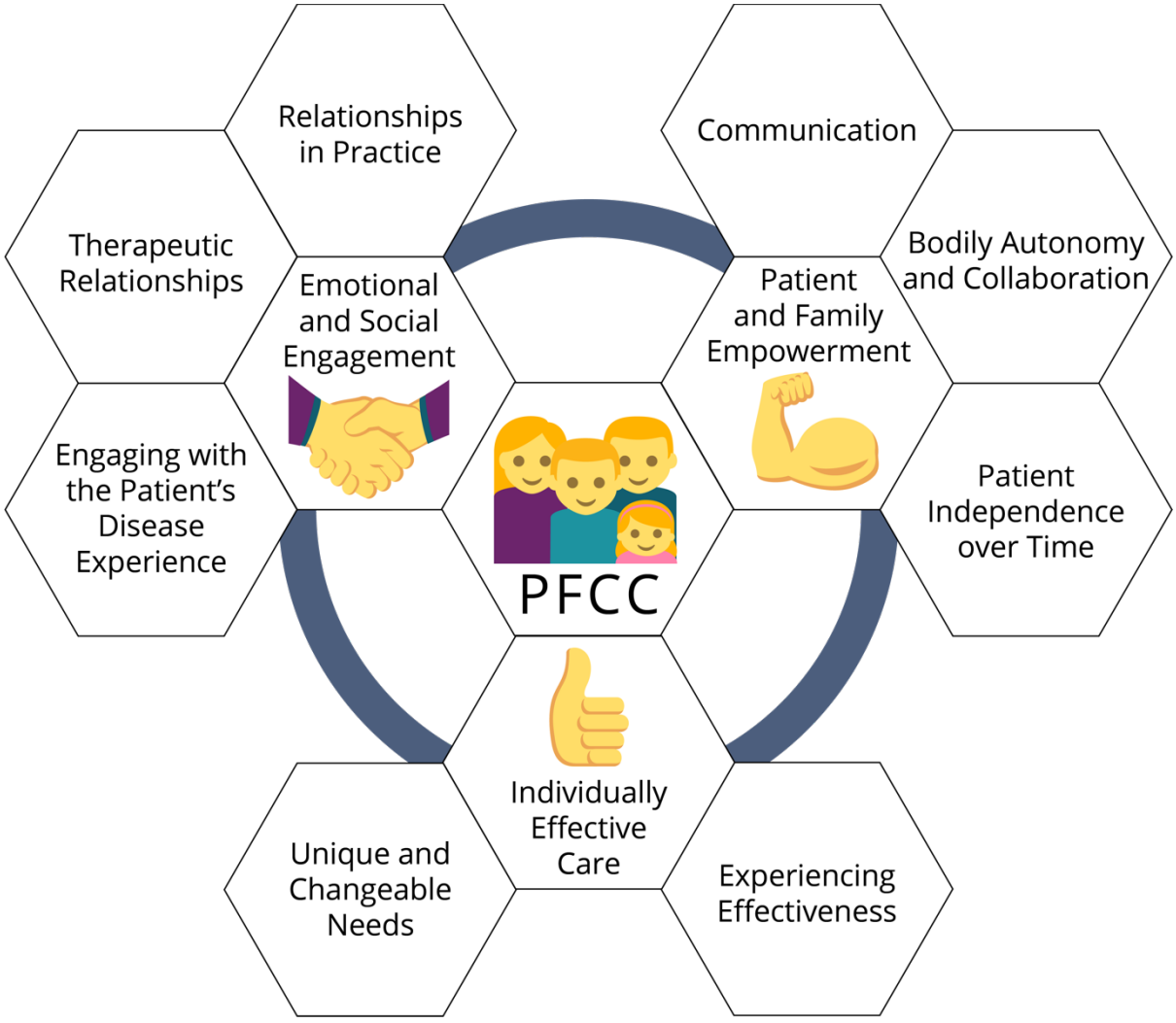

Figure 2.2. Characteristics and subthemes identified as being part of PFCC

Theme 1: Emotional and Social Engagement

Central to the care process for patients and families was a sense that the practitioner, patient, and family could interact with each other on an emotional and social level. In particular, they could develop therapeutic relationships that facilitated information sharing, self-management, and long-term engagement with treatment. Patients and families also suggested ways in which practitioners could enhance these relationships in practice. A close relationship helped practitioners to recognise and treat those aspects of the patient that were unique to their experience, rather than focusing purely on the disease process and symptoms.

Therapeutic Relationships. The therapeutic relationships between patients and families and practitioners were central to the experience of PFCC and were both a sort of "care capital" that facilitated and enhanced treatment, and a reflection of the perceived quality of the care received (Patterson & Lanier, 1999; Sly et al., 2014). Communication was enhanced when patients felt comfortable with, and trusted, their practitioners (Dovey-Pearce et al., 2005; Garvie et al., 2009; Grealish et al., 2013; Munson et al., 2012; Rudgley, 2013; Shaw et al., 2004; Sly et al., 2014). Trust could only be developed over time, with continuity of practitioners reported as an important facilitator of trust (Dovey-Pearce et al., 2005; Harper et al., 2014; Patterson & Lanier, 1999; Shaw et al., 2004).

> *They were there for me. . . . If it was just another program I wouldn't have honestly cared, I would have just disappeared. . . . But . . . they put the time and effort into trying to help me [and] all they ask from me is just to be better.* (Lucksted et al., 2015)

Patients and families emphasised their individual strengths and capabilities, particularly their expertise about the disease and its management (Grealish et al., 2013; Sasse et al., 2013; D. A. Stewart et al., 2001). Where practitioners did not recognise and value this experience, this could leave young people feeling that they were not being taken seriously

(Grealish et al., 2013; Lucksted et al., 2015). However, patients and families also acknowledged that as their experience changes over time, their understanding and ability to advocate for themselves would also change, requiring practitioners to be flexible in accommodating the patient and family's changing level of understanding over time (Delman et al., 2015; Doig et al., 2009; Harper et al., 2014; Rudgley, 2013).

Patients and families felt their individuality keenly and felt it important that practitioners treat them as they would any other person with whom they might come into contact, fostering a sense of normality (Offord et al., 2006; Patterson & Lanier, 1999; Rudgley, 2013). Stereotypes or negative assumptions held by practitioners based on the age of the patient or their disease left patients feeling like their practitioners did not care about them, which could disrupt the therapeutic relationship (Harper et al., 2014; Offord et al., 2006; Patterson & Lanier, 1999; D. A. Stewart et al., 2001).

The provision of emotional support, not just medical support, was an important part of the therapeutic relationship (Garvie et al., 2009; Gilmer et al., 2012; Offord et al., 2006; Patterson & Lanier, 1999; Rydström et al., 2013), and strongly supported the development of trustful relationships with practitioners (Delman et al., 2015; Grealish et al., 2013). Importantly, a lack of emotional support could lead to patients "burning out" over time, even when their care was medically appropriate (Fegran et al., 2014; Patterson & Lanier, 1999).

> *If I'm sad or feel alone, I would call my social worker for an appointment . . . we can meet and talk not only about the test results.* (Rydström et al., 2013)

The relationship between the patient and his or her family was also described as a source of support for young people (Lucksted et al., 2015; Rudgley, 2013). Treatment programmes that did not support this relationship left patients feeling further isolated and frustrated (Offord et al., 2006). Young people trusted their family members, particularly parents, to look out for them and to have their best interests at heart (Grealish et al., 2013).

Family members were affected by the patient's symptoms (Rudgley, 2013) and expressed a need for resources and support (Grealish et al., 2013). When appropriately supported, family members could help facilitate treatment success, helping patients make decisions and supporting them to engage fully with the health care process (Doig et al., 2009; Munson et al., 2012; Rudgley, 2013; Webster & Harrison, 2008).

> *My mom comes with me every time . . . I actually like her support . . . Having my mom come makes it feel less of a struggle.* (Lucksted et al., 2015)

Relationships in Practice. Patients and families reported that practitioners could foster relationships with them in a variety of ways. Patients and families highly valued practitioners who demonstrated a keen and ongoing interest in the patient and family and their wellbeing (Grealish et al., 2013; Munson et al., 2012; Rudgley, 2013; Sly et al., 2014). Listening attentively to patients and families to understand their perspective was strongly emphasised as valuable by patients and families, facilitating communication and enhancing treatment effectiveness (Delman et al., 2015; Dovey-Pearce et al., 2005; Grealish et al., 2013; Harper et al., 2014; Lee et al., 2006; Munson et al., 2012; Patterson & Lanier, 1999; Sly et al., 2014; Swift et al., 2013). Understanding the disease severity as experienced by the patient could also help to ensure that therapeutic messages were not seen as catastrophising or exaggerated (Nilson et al., 2012).

Conversely, practitioners who did not listen to, or consider, the concerns and opinions of patients and families could discourage patients from attending appointments (Nilson et al., 2012; Rudgley, 2013; Sly et al., 2014). Particularly effective were practitioners who created a relaxed and safe atmosphere, in which both parties could share personal stories, concerns, and experiences (Lee et al., 2006; Lucksted et al., 2015; Offord et al., 2006; Sly et al., 2014).

> *I'd look forward to our [weekly] sessions […] I knew I could keep going because soon we'd have key work and talk it through* (Sly et al., 2014)

Practitioners who made time to spend with patients and families were very positively regarded (Darrah et al., 2002; Fegran et al., 2014; Lucksted et al., 2015; Swift et al., 2013), particularly when they made the effort to engage outside scheduled appointment times or outside their office (Lee et al., 2006; Lucksted et al., 2015). Many patients felt that their practitioners were very busy, and did not want to waste their time, and so time spent was highly valued as a result (Fegran et al., 2014; Lee et al., 2006; Rudgley, 2013).

> *They would make the effort, and I like that. Instead of waiting for me to come to them, they would come to me, call me, ask me what's wrong you know … So now I like to come, and I look forward to talking with them.* (Lucksted et al., 2015)

Engaging with the Patient's Disease Experience. The experience of chronic disease was complex and difficult for many young people, and the nature of disease could leave young people feeling that their lives had become stagnant as they had been unable to engage with everyday life (Offord et al., 2006; Rudgley, 2013). Notably, the effect of disease on social networks was an important part of the disease experience, and patients and families described effective treatment as that which supported the maintenance of existing social networks and the development of new ones (Grealish et al., 2013; D. A. Stewart et al., 2001). Comparing their own experiences against those of their healthy peers could leave young patients feeling abnormal (Dovey-Pearce et al., 2005; Rydström et al., 2013), further isolating them from their peers and leaving them feeling lonely (Munson et al., 2012; Rudgley, 2013).

> *but I was very aware that they were getting on with their lives, erm, they were doing their 'A' levels, they were gonna be going off to university at the end of the year, and that was really hard for me cos I had fears of everybody going . . . and I'd never catch up . . . it meant that I sort of stayed stuck* (Offord et al., 2006)

This sense of abnormality and loneliness could leave patients with a range of complex emotions, including fear of relapse (Munson et al., 2012) and grief (Dovey-Pearce et al.,

2005). This could lead to denial of the disease and its effects (Nilson et al., 2012; Rudgley, 2013), to re-establish a personal sense of being "normal" (Nilson et al., 2012). Failure on the part of practitioners to engage with these emotions could hamper the ability of the patient and practitioner to work together to facilitate treatment (Fegran et al., 2014; Patterson & Lanier, 1999). Conversely, where particularly strong emotions related to the disease experience interfered with the young person's ability to engage with treatment, more directive treatment approaches were considered appropriate to allow the young person to become more comfortable and self-sufficient (Fegran et al., 2014; Offord et al., 2006).

Patients stressed how important it was for practitioners to engage with all facets of their experience of their disease, addressing its effects on their lives holistically rather than focusing narrowly on the pathology and treatment. Patients have multi-faceted lives (Lucksted et al., 2015; Munson et al., 2012) for a variety of non-medical reasons, and factors including housing needs (Gilmer et al., 2012; Patterson & Lanier, 1999), employment (Rudgley, 2013), and lack of a daily routine (Garvie et al., 2009) could all interfere with their ability to participate in treatment. To this end, patients valued providers who recognised the impacts that treatment could have on everyday life and assisted them to minimise these impacts (Garvie et al., 2009; Munson et al., 2012), and who supported them through the health care and social systems that they had to deal with.

> *Right now, I do need professional help . . . and the thing that's stopping me is basically time. I was going to school full-time, then I have to come home and take care of my daughter. So it's just a battle between when do I take the time to do it?* (Munson et al., 2012)

Patients and families felt that their personal experiences of disease gave them expertise that was fundamentally different from practitioners' medical expertise. Young people felt that they became experts on their own care, and felt that this expertise could

supersede the opinions of the medical team where they disagreed with their opinions (Nilson et al., 2012; Patterson & Lanier, 1999). Patients and families also strongly valued the experiences of peers who had the same health conditions, as emotional supports (Fegran et al., 2014; Gilmer et al., 2012; Offord et al., 2006; Patterson & Lanier, 1999; Rudgley, 2013) or as mentors (Patterson & Lanier, 1999; Shaw et al., 2004), noting that they could guide them through the experience in ways that their practitioners could not.

> *If they had somebody that they could talk to that's their own age that is going through some of the issues that they're going through, you know, I think that'd be really powerful.* (Gilmer et al., 2012)

Theme 2: Patient and Family Empowerment

Patients and families felt strongly that part of the role of practitioners was to enable and empower them to engage in care collaboratively with practitioners, rather than as passive recipients of medical expertise. The quality of communication between patients, families, and practitioners was the primary enabler of this ability for patients and families to collaborate, with poor communication by practitioners disempowering patients and families. The nature and level of this collaboration and communication was not stable over time, with patients and parents recognising the need for increased autonomy and independence with increasing age.

Bodily Autonomy and Collaboration. Patients repeatedly emphasised the importance of recognising ownership of their bodies (Shaw et al., 2004) and power in their own lives (Grealish et al., 2013). Information, both medical and service-based, was seen as strongly influencing patients' sense of control over themselves and their care. In particular, the rights to "know what's going on with your body" (Patterson & Lanier, 1999), to initiate help-seeking (Grealish et al., 2013), and to determine to whom that information was disclosed (Rydström et al., 2013) were deeply important to patients. Relevant medical information communicated clearly and at an appropriate level of complexity was highly

valued by patients and families (Dovey-Pearce et al., 2005; Grealish et al., 2013), as it helped patients and families better understand and predict disease (Rudgley, 2013). Patients could also use this information, given appropriate freedom by their practitioners (Grealish et al., 2013), to develop effective self-management strategies (Grealish et al., 2013; Rudgley, 2013).

Even with full and frank medical knowledge, however, patients and families were often unaware of available services (Darrah et al., 2002; Munson et al., 2012), in some cases learning about health services via serendipitous encounters with other professionals (Munson et al., 2012). This lack of knowledge about available services stemmed from difficulties communicating with practitioners (Rudgley, 2013), often because patients and families "don't know what they don't know" (Darrah et al., 2002).

> *The services are there. Sometimes you have to ask specifically. Like they don't just sort of say 'well these are the services that are out there for you.' You have to say 'I want this'. And then they'll tell.* (Darrah et al., 2002)

Patients and families expressed a strong desire for collaboration with practitioners (Offord et al., 2006) in which they could discuss their options and the potential benefits of those options (Grealish et al., 2013; Patterson & Lanier, 1999), ask questions (Rudgley, 2013), take time to consider the information (Delman et al., 2015), and then make decisions for themselves (Fegran et al., 2014; Grealish et al., 2013; Offord et al., 2006; Rudgley, 2013). Care that was collaboratively determined was valued by patients and families (Offord et al., 2006), and the resulting feeling of empowerment helped patients feel more in control of their own disease (Grealish et al., 2013) and improved their motivation and engagement with treatment (Doig et al., 2009; Fegran et al., 2014; Lucksted et al., 2015; Sly et al., 2014)

Communication. Families reported that they often had trouble understanding practitioners, and felt that being clearly understood was part of the practitioners' role (Darrah

et al., 2002). In particular, staff making decisions without close involvement of the patient and family could leave them feeling confused and frustrated (Offord et al., 2006; Rudgley, 2013), disengaged with treatment (Lucksted et al., 2015; Patterson & Lanier, 1999) and powerless (Offord et al., 2006). In addition, by failing to proactively inform and include patients, practitioners left patients out of forward planning and decision-making, leading some to believe that none had been done at all (Patterson & Lanier, 1999; Rudgley, 2013; Shaw et al., 2004).

> *I don't think my doctor thought about it. There were a lot of things that I didn't know or didn't think about, and I kind of went through things blind.* (Patterson & Lanier, 1999)

Difficulties communicating with practitioners were compounded by fear on the part of patients and families to speak up to practitioners. In some cases, this was because they did not know that they could assert themselves (Delman et al., 2015), or because they feared that by asserting themselves they would then have to take sole responsibility for their own care and lose the supports that they had in the past (Rudgley, 2013). Where practitioners gave patients more independence over time, they developed confidence and were better able to self-advocate (Delman et al., 2015; Rudgley, 2013), although failure to assert or manage newfound power could erode confidence very quickly (Offord et al., 2006; Shaw et al., 2004).

Patient Independence over Time. The parents' role in the care experience for the young adult was multifaceted, and changed over time. As the primary drivers of their children's care over the long term, parents often felt a sense of insight into their children, with mothers specifically feeling that they would recognise symptoms and concerns before clinicians did (Doig et al., 2009; Rudgley, 2013). As a result, they felt that they had a right to be centrally involved in their young adult children's care (Sasse et al., 2013), and a

responsibility to ensure that their children were protected from substandard care (Sasse et al., 2013; Shaw et al., 2004), unscrupulous providers (Saaltink et al., 2012), and immature decision-making (Sasse et al., 2013).

However, parents recognised the importance of supporting their children to develop independence and self-management, encouraging them to see providers alone and engage in appointments (Saaltink et al., 2012; Sasse et al., 2013; Shaw et al., 2004). Parents recognised that they may not know everything about their children, and trusted children and practitioners to share information and work in their adult child's best interests (Sasse et al., 2013).

> *The whole role for me of being a parent is to get them to that independent stage where they can think for themselves and do for themselves and be able to start to relate to other people in all aspects of their life.* (Sasse et al., 2013)

While parents recognised the need to assist their young adult children in developing independence in the health care process, young people needed practitioner support to achieve this in practice. They wanted, in particular, to be able to see their practitioners alone (Harper et al., 2014; Shaw et al., 2004), and gain access to information (Shaw et al., 2004). However, they had trouble telling their parents this (Shaw et al., 2004), especially in situations where their parents had "trouble letting go" (Fegran et al., 2014; Patterson & Lanier, 1999). In extreme cases, extreme parental involvement was identified as a potential barrier to treatment, dissuading patients from treatment (Lucksted et al., 2015; Webster & Harrison, 2008). Clinicians had an important role in facilitating young people's involvement in consultations (Dovey-Pearce et al., 2005), although this could be as simple as addressing the patient directly, rather than the parents (Shaw et al., 2004).

Theme 3: Individually Effective Care

Fundamental to the experience of PFCC was the requirement that the care be experienced as effective in achieving patients' and families' goals. Effective care was defined

as care in which the individualised needs of patients and their families were addressed in a way that they felt worked for them. Patients needed to be able to easily access experienced and knowledgeable professionals, as care delivered by practitioners who were not available or not perceived as skilful was not perceived as effective.

Unique and Changing Needs. Patients often emphasised that their needs were unique (Dovey-Pearce et al., 2005; Gilmer et al., 2012), dependent on the life and goals of the person themselves (Lucksted et al., 2015), and that this required flexibility on the part of the practitioner (Dovey-Pearce et al., 2005). Patients and families wanted to discuss the approach to care (Doig et al., 2009) and treatment methods (Rudgley, 2013) so that decisions could be tailored to their particular circumstances.

> *If you leave it up to the individual to pick goals or things that are essentially problems for them and they are working towards that, they can see the benefit of their improvements, and obviously they're a lot more satisfied with that.* (Doig et al., 2009)

In particular, the use of medication as a first resort was considered a warning sign that the care team did not really understand what the problems were (Lee et al., 2006). Young people held complex attitudes towards medication (Lee et al., 2006; Rudgley, 2013), although they were more likely to accept it if they were involved in the decision-making process (Lucksted et al., 2015).

Young people's needs change over time (Garvie et al., 2009), and this was particularly important in the context of transition from paediatric to adult services. There was a sense that services were taken away as young people got older (Munson et al., 2012; D. A. Stewart et al., 2001) without regard for the needs of the patient, or that where services were provided they were tailored towards younger children (Shaw et al., 2004). Patients and family members felt that individual needs and capabilities were a more important criterion for

transition than age (Fegran et al., 2014; Harper et al., 2014; Rudgley, 2013; Shaw et al., 2004; Swift et al., 2013).

> *it's not about the age. I don't believe anything is about the age. He (Psychiatrist) looked at it (referral) with other people and said you know where do you think, who would be best for her? . . . I think that would be better for people to do that rather than put them in a category because of their age because I don't think that's fair. We need services based on our needs not our bloody age* (Harper et al., 2014)

Addressing those needs of the patient and family that mattered to them led to more obvious benefits to the young person and their family members. Patients preferred treatment that they felt worked and from which they could see benefits (Doig et al., 2009; Gilmer et al., 2012; Lee et al., 2006), and engaged more strongly with those services that they felt helped them (Lucksted et al., 2015; Munson et al., 2012). Conversely, they were dissuaded from services that they did not feel helped (Lee et al., 2006), and stopped accessing those services or treatments that they felt did not benefit them (Lucksted et al., 2015; Munson et al., 2012)

Experiencing Effectiveness. For care to be experienced by patients and families as effective, they had to be able to access it consistently and easily accessible. Consistent care over the long term, particularly as young people got older and left paediatric services, was a major concern (Rudgley, 2013; D. A. Stewart et al., 2001; Swift et al., 2013). However, flexibility in appointment availability (Garvie et al., 2009) and access to services outside standard appointment structures (Swift et al., 2013) were also important for accessing more immediate care. Patients identified multiple barriers to seeing their practitioners, including long wait times, a lack of insurance, reliance on public transportation (Munson et al., 2012).

Patients and families wanted to deal with professionals who demonstrated their knowledge and skill (Lee et al., 2006), who were experienced in working with young people (Darrah et al., 2002; Dovey-Pearce et al., 2005), and who had some understanding of the

patient's health condition (Harper et al., 2014; Rudgley, 2013). They acknowledged that practitioners (in particular General Practitioners) may not have these skills (Grealish et al., 2013) and would rather be referred than seen by somebody who did not have the appropriate training and expertise (Garvie et al., 2009). Unprofessional conduct (Lee et al., 2006) and inconsistent information (Rudgley, 2013) could lead to reduced faith in the practitioner, which could lead to parents in particular not trusting them with their child (Sasse et al., 2013).

Discussion

The aim of the present review was to generate an understanding of PFCC that identifies and addresses the needs of young adults and their families by bringing together reports of studies that ask young people and their families living with a range of disease aetiologies how they define PFCC across a variety of care settings. The themes of the review, Emotional and Social Engagement, Patient and Family Empowerment, and Individually Effective Care, reflect the topics identified by a wide range of young people living with chronic disease and their families. While these themes are necessarily interdependent, they reflect the broad complexity of what young people and their families want from chronic disease management.

The development of a Therapeutic Relationship between practitioner and patient, driven by the practitioner's recognition of the patient's unique disease experience, is a powerful facilitator of the other aspects of PFCC. By recognising the importance of relationships as a kind of "care capital" with an inherent value to the health care delivery process, practitioners could make the health care interaction about more than just the disease and more about the person and their experience. Then, by demonstrating interest in the patient and their concerns, practitioners set the health care interaction up for success into the future.

Empowerment of Patients and Families was a feature of all stages of disease management, from the initial recognition of patients' personal autonomy by including them in decision-making to the gradual transition of control to patients from their parents and other caregivers over time. By encouraging collaboration through welcoming and encouraging active communication, practitioners bring young people and their families into the health care process. This, coupled with strong social and interpersonal relationships, enhanced communication through a sense of trust and support, allowing patients to establish themselves as legitimately part of the health care team rather than purely as the subjects of health care intervention.

Once these foundations of strong emotional and social engagement and patient and family empowerment were laid down, patients and practitioners could work towards addressing patients' individual needs. Once practitioners saw the unique and changing nature of the needs of their patients and their families, they were in a powerful position to assist them in working towards fulfilling those needs. By assisting patients to experience the achievement of their goals directly and personally, practitioners could demonstrate the effectiveness of management, encouraging engagement on an ongoing basis and helping to ward off "burnout". This experience of success also may help foster a sense of hope for the future, a component of patient-centred care particular to chronic disease settings (Hudon et al., 2012).

The essential components of PFCC for young adults with chronic disease suggested by the present study are quite similar to other extant models of patient-centred care in the published literature. In particular, the aspect of engagement between patient and family and practitioner is reflected in Mead and Bower (2000) in the themes of "patient-as-person", "doctor-as-person", and "the therapeutic alliance". The importance of an honest and open relationship in which both parties can act as what Mead calls "experiencing individuals" who

interpersonally influence each other is reflected in the depth with which patients and families discussed their experiences.

This may be contrasted with the model of patient-centred care developed by Kitson et al. in acute care settings (2013) , where the "relationship between the patient and the health professional" is described as "genuine", and facilitated by "open communication of knowledge, personal expertise, and clinical expertise". In their model, while a genuine relationship between the patient and practitioner is highlighted, there is much less emphasis on the practitioner as an emotional and social actor in the health care exchange. This contrasts with the strong emphasis in the present results on the continuity of practitioners over the long term, and the development of social relationships between practitioners and patients and family. The ongoing nature of the partnership between patient and practitioner, and thus the nature of the practitioner as an experiencing individual has been identified by Hudon et al. as being more important in chronic disease settings than acute settings (Hudon et al., 2012), as the relationship between patient and practitioner is extended over months or years, which may explain why this receives less weight in Kitson's study of acute care settings (2013).

Similarly, the theme of patient and family empowerment reflects Mead's (2000) focus on "sharing power and responsibility" and Kitson's (2013) concept of the "patient participating as a respected and autonomous individual", as well as their recognition of the personal expertise of the patient and the importance of open communication of knowledge. Again, the direct empowerment of the patient to deliver care and support their own health is not as strong in Kitson's (2013) model, which was also identified by Hudon et al. (Hudon et al., 2012) as more prominent in chronic disease settings than acute settings.

The present results also fit well with models of patient-centred care developed for other chronic diseases, for example, hearing loss. Grenness et al. (2014) studied a group of

older adults living with hearing loss, and highlighted the importance of addressing the unique and changeable needs of the patient and their family and the importance of the therapeutic relationship, as well as the importance of practitioner professionalism and having more than merely technical skill. Grenness also highlighted the provision of relevant information in an honest and complete manner as an important element of patient control and efficacy, similar to the present results. However, as might be expected due to the older age of their participants, Grenness' respondents did not experience the same kinds of tensions with family members that were reported in the present study's literature, and so this is not reflected in their model.

Epley et al. reported five key elements of FCC present across the literature: family choice, family strengths, family–professional relationship, individualised family services, and the family as the unit of attention (Epley et al., 2010). The importance of family choice is reflected in the sub-theme "Bodily Autonomy and Collaboration" in young people's focus on their right to make their own decisions and lead their own care. Epley's identification of individualised services being important to FCC was also reflected by young adults in the literature, with services only being experienced as effective if they addressed patients' unique needs. The family-professional relationship was highlighted by young adults, with them highlighting not only the importance of the therapeutic relationship but also how to achieve these "relationships in practice" - in particular, recognising their individual strengths and abilities to lead and manage their own care. Consideration of Epley's concept of the family as the unit of attention considering the present results reveals an interesting tension; the role of parents in the health care process was not clear, with parents acknowledging that they had to work to step back and allow their children additional agency and autonomy over time.

In contrast, there are significant differences between the present results and the Neurodevelopmental Clinical Research Unit framework for family-centred care (FCC)

developed by Rosenbaum et al. (2009). This model highlights the parents as the unit of agency within the family, rather than the family as a support to a child able to make decisions. This model has been developed in a child rehabilitation context, and so focuses on the experiences and needs of children, rather than those of young adults. Children have yet to develop the personal autonomy and capabilities that are characteristic of what Arnett called "emerging adulthood", and so models of family-centred care that focus on young children may be unable to incorporate these capabilities into their structure.

This problem is compounded by the populations studied by the papers identified in this systematic review, of which only seven incorporated the views of parents, only one included siblings, and none incorporated other family members such as grandparents, partners, or children. The role of parents in facilitating their children's eventual independence and success was a feature of those reports that included them in data collection and analysis, and this seems to be an important part of patient- and family-centred care in this population. Further research in this area should incorporate the experiences and opinions of parents and other family members to be sure that this important facilitation role is being enhanced as control of the clinical process passes from them to their young adult child.

Papers identified in the present review also focused strongly on the experience of transition from paediatric to adult services. This is a time of significant change for young people living with chronic diseases, and the findings of this review suggest that young people and their families desire stability and continuity in their interactions with practitioners. As young people engage with models of care that require them to transition between services and service providers, they may focus on this lack of stability, de-emphasising other areas of importance in the interest of addressing the primary threat to their sense of PFCC. Further investigation in settings where transition is not a feature would be helpful in elucidating

additional features of PFCC that may emerge when existing practitioner relationships are not under threat.

The present review of qualitative studies is, necessarily, two steps removed from the experiences of the young people living with chronic health conditions whose voices have been quoted from other studies and used as data in the current study. While the research team have attempted to focus on the words of participants in the synthesis process, identified findings have been shaped by the decisions of the individual researchers who conducted these component studies, and further interpreted by the current research team. This process of repeated interpretation, having been conducted by health researchers runs the risk of influencing the development of concepts already familiar to health researchers - for example, shared decision-making, patient involvement, and the therapeutic relationship (Larkin & Thompson, 2012, p. 112). As a result, reviews such as this should not be assumed to reflect the wishes and needs of young adult patients in every care setting. Rather, directly engaging with patients and working with them to co-design services and service improvements is vital (Bate & Robert, 2007).

The findings of this study highlight actions that health care practitioners could take to encourage PFCC in their everyday practice, as seen through the eyes of young people. In this way, they complement the more theoretical framework put forward by Stewart et al. (M. Stewart et al., 2014) that suggests a way of conceptualising health care and disease to enable patient-centred care. The present results suggest three immediate measures of patient- and family-centredness that may be useful for clinicians as part of a reflective practice methodology: (1) Did I engage emotionally with my patient and their family on an honest level; (2) Did I empower the patient and their family to participate in decision-making and health-care delivery; (3) Did I focus care on the goals of the patient and family as they see them? In this way, practitioners may be more able to assess their own practice to better

ensure that they are delivering care to their young adult patients in a patient- and family-centred fashion.

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CHAPTER 3 – EFFECTS OF PATIENT- AND FAMILY-CENTRED CARE FOR YOUNG ADULTS WITH CHRONIC HEALTH CONDITIONS AND THEIR FAMILY MEMBERS: A SYSTEMATIC REVIEW (ACCEPTED FOR PUBLICATION)

Rationale

After defining a model of PFCC in young adults living with chronic disease as described in Chapter 2, it followed to ask whether efforts to move towards this model are effective in improving aspects of care. To address this question, it was originally intended to use the same dataset as the previous chapter, but target the analysis towards the impacts of patient- and family-centred approaches. It was hoped that this would allow a relatively straightforward way to validate the model, and determine whether or not it is associated with improved outcomes, similar to work seen in other care settings and disease aetiologies.

However, when this was attempted, it was found that the data collected in the previous review of the literature did not adequately address the new question, and so an additional review had to be conducted. As the model identified in the first systematic review was new, and at that time unpublished, no studies testing it in its entirety existed. Therefore, its effectiveness at improving the delivery of care could not be appraised. In an attempt to determine whether implementation of this model in practice would be likely to improve care for young people with chronic health conditions, the decision was made to explore the impacts of *Patient- and Family- Centred Interventions* (PFCIs) – changes to the way in which care is delivered that are an attempt to move towards any of the principles outlined in the model.

There was relatively little data available in the peer-reviewed literature, and that which was available covered a wide variety of approaches, outcome measures, and effect

sizes. A mixed-methodology systematic review of this literature resulted in to the following paper, which has been accepted for publication by the journal Emerging Adulthood. The paper is presented in its entirety, with minor formatting changes to ensure consistency with the remainder of the thesis.

While, like Chapter 2, this chapter deals with young adults with chronic diseases more broadly, rather than those with hearing loss in particular, this work was particularly useful when designing the study reported in the Chapter 4. The highly variable nature of the results of this review, and the lack of effect of PFCIs on the majority of measures of healthcare outcomes reported in the literature, highlight the importance of treating patient-centredness of care as an outcome in itself, separate from any potential effects on disease symptomatology or health-related quality of life. Rather, patient-centredness may have positive effects on patient satisfaction and empowerment, the improvement of which are noble goals in themselves. The presence of these effects support the inclusion of broad outcome measures including those focusing on the patient experience when assessing the effectiveness of patient-centred care in practice.

Introduction

Patient- and family-centred approaches to healthcare are increasingly being adopted by Government and non-Government organisations alike as a more effective way of delivering healthcare than traditional models of care, which prioritise diagnosis and cure of the health condition over addressing patient concerns (Jo Delaney, 2017). This movement is supported by research evidence demonstrating that patient- and family-centred care (PFCC) improves patient and healthcare outcomes across a range of pathologies and health conditions, including decreased morbidity (Kuo, Bird, & Tilford, 2011), lower mortality (Glickman et al., 2010; Meterko, Wright, Lin, Lowy, & Cleary, 2010), improved patient satisfaction (Beach et al., 2005; Jakimowicz, Stirling, & Duddle, 2015), increased adherence

to treatment (Arbuthnott & Sharpe, 2009; Beach et al., 2005; Blackwell, 1996; Coe, Prendergast, & Psathas, 1984; Garrity, 1981; Glickman et al., 2010), and decreased cost of care (Kuo et al., 2011; Stewart, Ryan, & Bodea, 2011). These benefits are particularly important for patients living with chronic health conditions (Bokhour et al., 2009), as their partnership with their health care team extends over a long period (Hudon et al., 2012). In addition to these demonstrated patient and healthcare outcomes, importantly, patients have expressed a preference for patient-centred approaches (Rademakers, Delnoij, Nijman, & de Boer, 2012) with a strong therapeutic relationship between patient and practitioner (Grenness, Hickson, Laplante-Levesque, & Davidson, 2014) facilitating communication and partnership (Little et al., 2001).

The appropriate management of chronic health conditions is particularly important for young adults (defined here as those aged between 16 and 25 years, in order to capture those young people leaving secondary education) who are likely to be undergoing significant change in many aspects of their lives, including moving away from parents, establishing careers of their own, developing romantic relationships, and having children (National Centre for Vocational Education Research Ltd, 2014). This period of life, sometimes termed "emerging adulthood", is distinct from adolescence or adulthood because of the pace, extent, and variety of personal change (Arnett, 2000). Arnett's theory of emerging adulthood focuses strongly on the development of personal identity, which is closely linked with the development of a sense of control over one's own body and life course, known as *agency* (Schwartz, 2005). Within healthcare interactions, patient agency is particularly important, as the empowerment of patients within the healthcare process can lead to greater patient satisfaction and treatment adherence (Loukanova, Molnar, & Bridges, 2007).

Decisions made early in life regarding the management of chronic health conditions can potentially have long-lasting effects for the remainder of a young person's life.

Challenges to the effective management of individuals with chronic conditions, such as the requirement to transition between paediatric and adult health services, can put significant strain on young people (Fegran, Hall, Uhrenfeldt, Aagaard, & Ludvigsen, 2014). Their long history of medical management can also lead to "burnout" and subsequent disengagement with the healthcare process (Patterson & Lanier, 1999). As a result, effective management of chronic health conditions and the maintenance of patient engagement during emerging adulthood is particularly important. By developing the ability of patients to direct the course of their own treatment and management, PFCC has the potential to improve engagement with care, with subsequent benefits for outcomes (Stewart et al., 2014, pp. 349-350).

Illness, particularly where that illness is life threatening, can also have significant traumatic effects on other family members. This trauma is particularly a problem for the parents (Landolt, Vollrath, Ribi, Gnehm, & Sennhauser, 2003; Noyes, 1999) and siblings (Kaplan, Kaal, Bradley, & Alderfer, 2013) of young children with chronic health conditions. The effects of health conditions on other family members can lead to reduced outcomes for the person with the chronic health condition, through reducing their ability to support their family member, assist them with decision-making, or through increasing stress in the home (Hickman & Douglas, 2010). Being listened to by care staff, and subsequently developing relationships with them, has been described by mothers as a way of reducing this trauma and facilitating parental coping (Noyes, 1999). By incorporating family needs, views, and preferences, PFCC may therefore improve the experience of family members and reduce distress (Brown, Mace, Dietrich, Knazik, & Schamban, 2015). To date, little has been reported on the extent of these effects among family members of young adults living with chronic health conditions, or on whether interventions aimed at improving the patient- and family-centredness of care improve their experiences of care.

In our previous systematic review of the literature, we demonstrated that PFCC for young adults living with chronic health conditions comprises three main factors: (1) congruent interpersonal engagement between the patient and family and practitioner; (2) the empowerment of the patient and their family; and (3) the pursuit of effectiveness as defined and experienced by the patient and family themselves (Allen, Scarinci, & Hickson, 2018). Each of these factors may be addressed by interventions targeting aspects of the care experience, such as the nature of patient-practitioner communication itself, the information provided to patients, or the introduction of structured, patient-led goal setting. Importantly, the development of PFCC in a health system is not a single achievable event, but rather a journey towards a more patient- and family-centred approach, where individual parts of the care process are modified to better incorporate the views and beliefs of patients and their families (Institute For Patient- and Family-Centered Care, 2017). As a result, interventions targeting these individual factors may be seen as moves towards PFCC in a health system.

In order to improve the performance of health services and attempt to align service delivery with PFCC, there is a need for high-quality evidence of the effects of any proposed interventions. As this evidence is best delivered by systematic reviews of the literature with meta-analysis (Harris, 2006, p. 434), the present review builds on our previous work (Allen et al., 2018) to determine what effect patient- and family-centred interventions (PFCIs) have on the effectiveness of care, as defined by both objective external measures and subjectively by young adult patients and families themselves. PFCIs are here defined as changes to the delivery of care targeting any one of the three individual factors of PFCC described above. The research question of this review was "What are the impacts of PFCIs on the effectiveness of health care provided to young adults living with chronic health conditions?"

Method

The protocol for this systematic review has not been previously published.

The appraisal and inclusion of mixed-methods research in systematic reviews is controversial, with most systematic reviews of the literature focusing on reports of either quantitative or qualitative studies. However, restricting reviews based on method can reduce the clinical applicability of the results (Joanna Briggs Institute, 2014), especially in cases where extant literature is of diverse methodologies and approaches. In these cases, segregated synthesis designs, where quantitative and qualitative reports are synthesised separately prior to being integrated, can allow for actionable quantitative findings to be complemented by qualitative inquiry (Sandelowski, Voils, & Barroso, 2006). Due to the variety of methodological approaches evident in the literature addressing the research question, a segregated design mixed-methods review was conducted in the present study.

Data Sources

A range of search strategies, detailed in Table 3.1, were used to identify literature for consideration for this review. Initially, CINAHL Terms and MeSH Headings relevant to young adults living with chronic health conditions were identified with the assistance of a health sciences librarian, which were then used to conduct searches in CINAHL Complete and MEDLINE. This initial pool of papers was then screened to identify any further relevant keywords, which were then used for searches in MEDLINE (via EBSCOHost), CINAHL Complete (via EBSCOHost), PsycINFO, and EMBASE. No date criterion was used for these searches, due to the long history of development of PFCC across a range of clinical specialities.

Table 3.1

Initial database search details

Database	Search Term	Date	Total
MEDLINE	(MH Adolescent OR MH Young Adult OR MH Transition to Adult Care) NOT (MH Aged OR MH Middle Aged) AND ((MH Professional-Patient Relations OR MH Professional-Family Relations OR MH Consumer Health Information OR MH Information Seeking Behavior OR MH Access to Information) OR (MH Patient Participation OR MH Patient Preference OR MH Personal Satisfaction OR MH Decision Making OR MH Health Communication OR MH Personal Autonomy OR MH Independent Living) OR (MH Needs Assessment OR MH Patient Care Planning OR MH Continuity of Patient Care) OR (MH Patient-Centered Care OR MH Family-Centered Care)) AND (LA eng AND (PT Clinical Trial OR PT Clinical Trial, Phase I OR PT Clinical Trial, Phase II OR PT Clinical Trial, Phase III OR PT Clinical Trial, Phase IV OR PT Comparative Study OR PT Controlled Clinical Trial OR PT Evaluation Studies OR PT Multicenter Study OR PT Randomized Controlled Trial OR PT Twin Study OR PT Validation Studies)	8/8/2017	2,885

Database	Search Term	Date	Total
EMBASE	([adolescent]/lim OR [young adult]/lim OR 'transition to adult care'/exp) NOT ('aged'/exp OR 'middle aged'/exp) AND ('professional-patient relationship'/exp OR 'doctor patient relation'/exp OR 'nurse patient relationship'/exp OR 'personal experience'/exp OR 'medical information'/exp OR 'decision making'/exp OR 'patient empowerment'/exp OR 'access to information'/exp OR 'patient autonomy'/exp OR 'personal autonomy'/exp OR 'patient participation'/exp OR 'independent living'/exp OR 'personal autonomy'/exp OR 'consumer health information'/exp OR 'information seeking'/exp OR 'goal setting'/exp OR 'goal attainment'/exp OR 'needs'/exp OR 'needs assessment'/exp OR 'holistic care'/exp OR 'family centered care'/exp) AND [humans]/lim AND [clinical study]/lim AND [english]/lim AND [article]/lim	17/8/2017	7,769
CINAHL	(MH "Adolescence" OR MH "Young Adult") NOT (MH "Aged" OR MH "Middle Age") AND ((MH "Professional-Family Relations" OR(MH "Professional-Client Relations" OR MH "Professional-Patient Relations" OR MH "Physician-Patient Relations" OR MH "Nurse-Patient Relations" OR MH "Truth Disclosure" OR MH "Communication Skills") OR (MH "Decision Making, Patient" OR MH "Empowerment" OR MH "Decision Making, Family" OR MH "Health Information" OR MH "Consumer Health Information" OR MH "Consumer Participation" OR MH "Health Services Accessibility" OR MH "Access to Information") OR (MH "Individualized Medicine" OR MH "Goal-Setting" OR MH "Goal Attainment") OR (MH "Patient Centered Care" OR MH "Family Centered Care")) Limiters: English Language, Research Article, Human	17/8/2017	4,541
Total from searches			15,195
Articles identified from authors' library		17/8/2017	15

Database	Search Term	Date	Total
Total before initial deduplication			15,210
Total after initial deduplication			14,208
Total after second deduplication			14,197

Initial Screening

After an initial deduplication process (Rathbone, Carter, Hoffmann, & Glasziou, 2015), 14,208 articles were loaded into a web-based screening tool for screening by the first author using titles and abstract (Ouzzani, Hammady, Fedorowicz, & Elmagarmid, 2016).

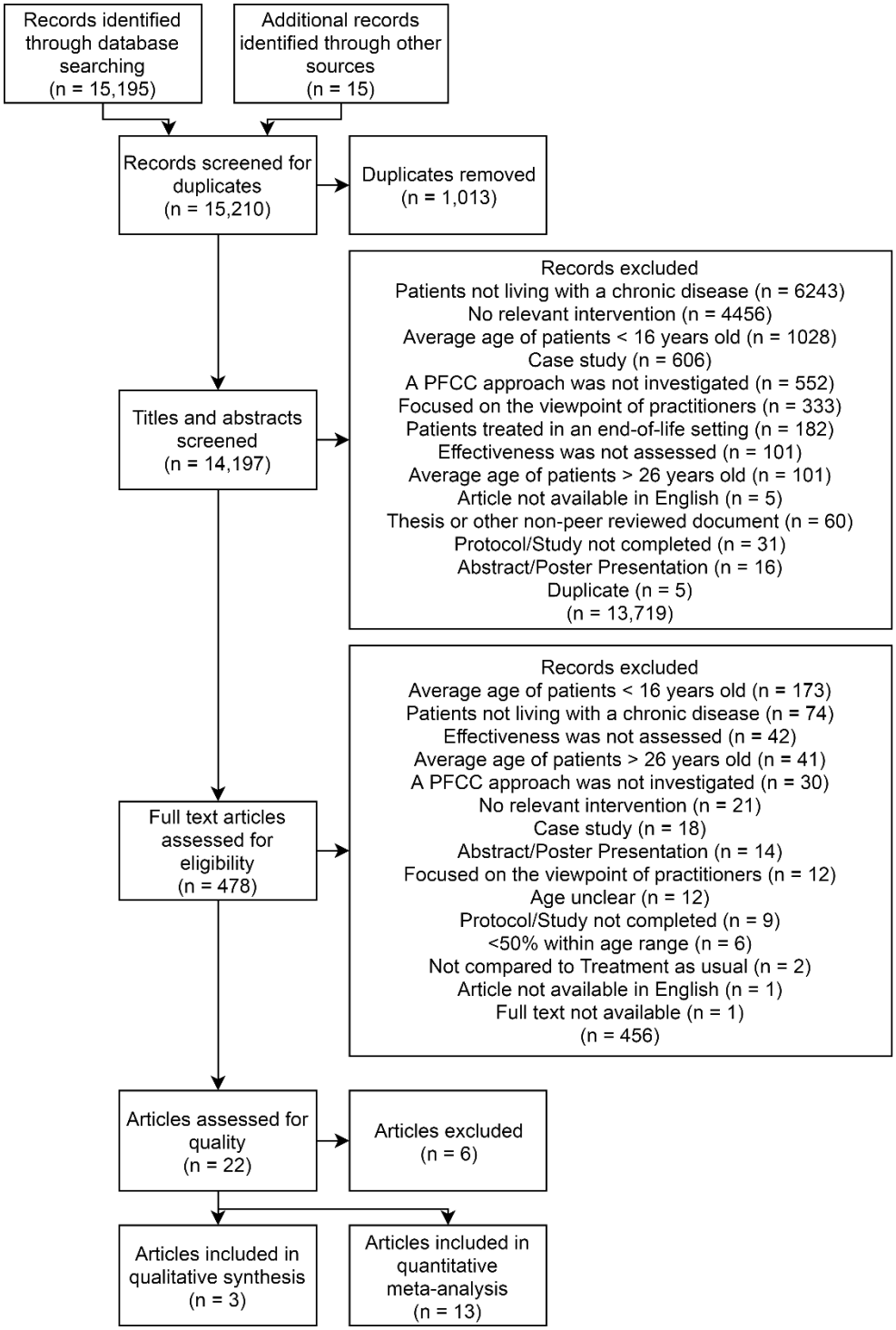


Figure 3.1. Flow chart for this review.

Each title and abstract was read and assessed against the following inclusion criteria by the first author:

a) Patients were living with a diagnosed chronic condition

b) At least 50% of subjects were between the ages of 16 and 25

c) A change to the way care was delivered was introduced (PFCI)
d) The results of this change were assessed relative to standard care
e) The change addressed one or more of the factors of PFCC: interpersonal engagement between the patient and family and practitioner, empowerment of the patient and family, or the pursuit of effectiveness as defined and experienced by the patient or family(Allen et al., 2018)
f) The results of the care were assessed relative to the patient and/or family (acceptability to practitioners was not considered, and where a qualitative approach was used, patient and family responses had to be separable from those of practitioners)
g) Article was available in English
h) The article was not a case study or multiple case studies
i) The patients were not receiving end-of-life care or planning
j) The report was peer-reviewed and available in full

For the purposes of this review, the distribution in age of participants was estimated using the reported mean and standard deviation, the median and interquartile range, or the age range if neither of these were reported. Algorithms used to determine whether papers satisfied the age criterion are presented in the appendix. Health conditions were categorised according to the International Classification of Diseases and Related Health Conditions, Tenth Revision (ICD-10, see World Health Organisation, 2016). The chronic nature of a health condition was determined by the Beta Chronic Condition Indicator for ICD-10-CM, which is a classification of ICD disease codes into either chronic or non-chronic, where a chronic condition is defined as “one that: lasts 12 months or longer and meets one or both of the following tests: (1) it places limitations on self-care, independent living, and social interactions; and (2) it results in the need for ongoing intervention with medical products,

services, and special equipment." (Healthcare Cost and Utilization Project (HCUP), 2017). Conditions related to pregnancy (O00-99), gender identity disorders (F64), obesity (E66), reactions to severe non-medical stress and trauma (F43), and the use of alcohol, tobacco, and other drugs (F10-19) were not considered for the purposes of this review, as treatment for these conditions was considered by the research team to be likely focused on removing or curing the underlying condition, distinct from that for other chronic health conditions. Help-seeking, such as for psychological counselling, was not considered evidence of a diagnosed health condition. Pre-post interventions where ongoing standard care prior to an intervention was compared to modified care were included where there was no additional change to care or the patient's disease progression over the course of the study (such as a major health care transition). During this process, a further 11 duplicates were identified and removed from consideration.

Of the 14,197 records that were initially screened, 143 (1%) were randomly chosen to be additionally independently screened by the second author according to the inclusion criteria. High agreement was obtained (98% agreement), with only three papers (2%) included by the first author that were excluded by the second author. All three of these papers were subsequently excluded by the first author on further screening.

Full-text Screening

After this initial screening process, 478 papers remained. The full text of each remaining paper was then obtained and the same inclusion criteria applied by the first author. After this process, 22 papers remained. These papers were separated by the first author based on their methodological approach into Qualitative (n = 8) and Quantitative (n = 14) papers. While several studies used both qualitative and quantitative techniques, none of these met the inclusion criteria across both approaches. Of these 478 papers, 48 (10%) were randomly selected for independent assessment by the second and third authors according to the

inclusion criteria. High agreement was obtained (96% agreement), with disagreement on only two papers (4%) which were included by the second author. These cases were discussed by the research team and were subsequently excluded.

Table 3.2

Critical appraisal checklists

Qualitative Study	**Quantitative Study**
1. Was there a clear statement of the aims of the research?	1. Did the study address a clearly focused issue?
2. Is a qualitative methodology appropriate?	2. Did the authors use an appropriate method to answer their question?
3. Was the research design appropriate to address the aims of the research?	3. Were the cases recruited in an acceptable way?
4. Was the recruitment strategy appropriate to the aims of the research?	4. Were the controls selected in an acceptable way?
5. Was the data collected in a way that addressed the research issue?	5. Was the exposure accurately measured to minimise bias?
6. Has the relationship between researcher and participants been adequately considered?	6. Was the outcome accurately measured to minimise bias?
7. Have ethical issues been taken into consideration?	7. Have the authors taken account of the potential confounding factors in the design or in their analysis?
8. Was the data analysis sufficiently rigorous?	8. Was the follow up of subjects complete enough? Was the follow up on subjects long enough?
9. Is there a clear statement of findings?	9. Have ethical issues been taken into consideration?
	10. Do you believe the results?

Quality Appraisal

The Qualitative and Quantitative quality appraisal tools developed from the Critical Appraisal Skills Programme tools in Allen et al. (2018) were applied to the relevant section of each paper by the first author and have been reproduced in Table 3.2. Where a paper used both qualitative and quantitative approaches, only the approach that passed the inclusion criteria was considered. A total of 13 Quantitative and 3 Qualitative papers remained

following critical appraisal. Of the 22 papers proceeding to quality appraisal, 9 (40%) were randomly selected and additionally independently assessed by the second author using the tools. Both authors agreed in all cases.

Among the 14 quantitative reports, one paper assigned participants to experimental or control group by attendance at a particular clinic, but it was unclear whether the two clinics were similar prior to the intervention, and so this study was excluded.

Table 3.3

Critical Appraisal Results for Quantitative Papers

Quantitative Papers	**Q1**	**Q2**	**Q3**	**Q4**	**Q5**	**Q6**	**Q7**	**Q8**	**Q9**	**Q10**	**Pass**
Godart et al. (2012)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Grawe et al. (2006)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Hagner et al. (2012)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Hammonds et al. (2015)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Jang et al. (2017)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Kaihin, Kasatpibal, Chitreechuer, and Grimes (2015)	Y	Y	Y	Y	Y	Y	N	Y	Y	Y	N
le Grange et al. (2007)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Levin and Rotheram-Fuller (2011)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Mackie et al. (2014)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
McCann et al. (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Mosher, Menn, and Matthew (1975)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sattoe et al. (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Schmidt et al. (2016)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Zaitsoff et al. (2008)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Note. Column headings refer to the question numbers as presented in Table 3.2. A Y is present where the authors determined that a question could be answered in the affirmative for a particular paper, and an N where it could not.

Of the eight qualitative papers, five were excluded, primarily because the relationship between the researcher and the participants was unclear, or because the data analysis was not in sufficient detail to justify the findings.

Table 3.4

Critical Appraisal results for Qualitative Papers

Qualitative Papers	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Pass
Gorter et al. (2015)	Y	Y	Y	Y	Y	Y	Y	N	Y	N
Brothers et al. (2014)	Y	Y	Y	Y	N	N	N	Y	Y	N
Pycroft, Wallis, Bigg, and Webster (2015)	Y	Y	Y	Y	Y	N	Y	N	N	N
Bobier et al. (2009)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sawin et al. (2015)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Quinn, Gleeson, and Nolan (2013)	Y	Y	N	Y	Y	N	Y	N	Y	N
Solórzano and Glassgold (2010)	Y	Y	Y	Y	Y	N	Y	Y	N	N
Price et al. (2011)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Note. Column headings refer to the question numbers as presented in Table 3.2. A Y is present where the authors determined that a question could be answered in the affirmative for a particular paper, and an N where it could not.

Quantitative Meta-Analysis

Estimated effect sizes were calculated for all findings within each study for which sufficient information was provided. Effect sizes for studies measuring continuous outcomes were calculated using the method described by Borenstein, Hedges, Higgins, and Rothstein (2009, pp. 26-28), as within-groups standard deviations were reported in all cases. For studies that reported binary data, log odds ratios were calculated (Borenstein et al., 2009, pp. 36-37) which were then converted to Hedges g using the correction proposed by Hasselblad and Hedges (1995). Calculations were performed in R (3.3.3), using the readxl (1.0.0), plyr (1.8.4) and metafor (2.0-0) packages.

All quantitative findings from the identified studies were pooled and then categorised by the authors into groups based on similarity of the aspect of effectiveness that was

measured, as determined inductively from the identified papers. In this case, three categories were identified as being useful for meta-analysis: 1) Disease severity, as measured by patient self-report, structured symptom questionnaire, or objective medical test; 2) Health-related Quality of Life (HRQoL); and 3) Self-Determination/Self-Efficacy. In addition, two studies reported measures of adherence to treatment, one study measured the presence or absence of disease, and one reported patient satisfaction, which are described below but excluded from meta-analysis. Random-effects models were used to estimate aggregate treatment effects, due to the varied interventions and outcome measures. Where multiple findings within a category related to the same study, a mean effect size (and mean standard error) weighted by the degrees of freedom was used. In particular, two papers were conducted within a single study on the same sample (although at two different time points) and so were combined in this way (le Grange, Crosby, Rathouz, & Leventhal, 2007; Zaitsoff, Doyle, Hoste, & le Grange, 2008). The proportion of participants falling within the identified age range was not used to weight effects as no consensus in the literature on how to address this problem could be found.

DFBETAs (a measure of the difference between parameters estimated with and without a particular observation) were calculated to determine likely outliers within each category (Viechtbauer & Cheung, 2010). The Trim and Fill method was used to identify studies likely missing due to publication bias (Duval & Tweedie, 2000). No likely publication bias was identified within any of the categories, with the number of missing studies in every case estimated as zero. Egger's Test was also used to detect asymmetry in the distribution of findings (Sterne & Egger, 2005). PET was evaluated for effect size estimation as suggested by the tool provided by Carter, Schönbrodt, Gervais, and Hilgard (2017, May 30). PET provided larger estimates of effect sizes in each final model (those with overly influential studies removed) than random-effects modelling, and so the more conservative estimates provided by random-effects modelling have been reported.

Qualitative Meta-Synthesis

Qualitative findings were synthesised using a modified version of the meta-aggregation methodology described by Lockwood, Munn, and Porritt (2015) Findings were identified and described along with units of supporting data. Where a verbatim statement of a finding could not be extracted, a modified finding was recorded that reflected the authors' understanding of that finding (see Allen et al., 2018). These findings were then sorted by the first author into categories and themes. This process was discussed by all members of the research team until consensus was reached.

The results of this meta-synthesis were then combined with the results of the quantitative meta-analysis following the segregated mixed research synthesis model described by Sandelowski et al. (2006). In this model, quantitative and qualitative results are considered complementary, and so, qualitative results can be used to develop a stronger understanding of why particular quantitative results have been obtained. To achieve this, quantitative meta-analysis and qualitative meta-synthesis are conducted separately, and the results from the meta-synthesis are compared with meta-analytic results to develop a deeper understanding of the topic under study.

Table 3.5

Extracted details of papers

	Disease	% in age range	ICD	Intervention	Aspect of PFCC	Outcome Category	Outcome	N	g	SEg
Godart et al. (2012)	Anorexia Nervosa	64.6	F50.0	Adjunctive Family Therapy	Family Empowerment	Disease Symptoms	Eating Disorder Inventory	30;29	0.0274	0.2618
						Disease Presence	BMI < 10th percentile	30;29	0.5977	0.3012
Grawe et al. (2006)	Schizophrenia	53.1	F20	Integrated early psychosis treatment	Family Empowerment	Disease Symptoms	Persistent Symptoms	30;20	-0.0472	0.2372
						Adherence	Good drug adherence	30;20	-0.0837	0.2106
						Adherence	Good therapy adherence	30;20	1.3675	0.6906
Hagner et al. (2012)	Autism Spectrum	100	F84.0	Family-Centred Transition Process	Empowerment/Effectiveness	Self-determination	Arc Self-determination Scale	15;17	3.4039	0.5537
Hammonds et al. (2015)	Depression	75.3	F33	Medication Reminding	Effectiveness (Accessibility)	Adherence	Drug Adherence	30;27	0.3769	0.1622
						Disease Symptoms	Beck Depression Inventory	30;27	0.3054	0.2668
Jang et al. (2017)	Irritable Bowel Syndrome - Constipation Predominant	98.1	K58.9	Cognitive-Behavioural Therapy	Empowerment	Disease Symptoms	GI Symptoms	23;20	2.7474	0.4257
						Disease Symptoms	Anxiety	23;20	4.0181	0.5303
						Disease Symptoms	Depression	23;20	4.0270	0.5311
						Disease Symptoms	Stress	23;20	3.0140	0.4462
Levin and Rotheram-Fuller (2011)	Visual Impairment	71.4	H54	Empowered Curriculum	Empowerment	Self-determination	AIR Self-determination Scale	13	-0.0444	0.3923
Mackie et al. (2014)	Congenital Heart Disease	68.8	Q20-Q24	Transition Readiness Intervention	Empowerment	Self-determination	TRAQ (Self-management)	27;31	0.4446	0.2665
McCann et al. (2013)	Psychosis	90.0	F20, F29	Family Bibliotherapy	Family Empowerment	Disease Symptoms	K10 Score	53;52	1.5358	0.2044
						HRQoL	SF-12 Physical	53;52	2.6500	0.2461
						HRQoL	SF-12 Mental	53;52	2.6727	0.2471
Mosher et al. (1975)	Schizophrenia	62.5	F20	Patient-centred residential care	Engagement/Effectiveness	Disease Symptoms	Global Psychopathology	10;8	1.1576	0.5121
Sattoe et al. (2013)	End-stage Renal Disease	90.0	N18.6	Camp COOL	Engagement/Effectiveness	Self-determination	General self-efficacy	32	0.2913	0.5046
						HRQoL	HRQoL	32	0.0189	0.2500
Schmidt et al. (2016)	Type I Diabetes	67.7	E10	Transition-oriented patient education	Empowerment	Self-determination	General self-efficacy	274	0.2611	0.1236
						Satisfaction	CHC-SUN	274	0.1480	0.1233
						HRQoL	EUROHIS QOL-8	274	-0.0504	0.1231
Zaitsoff et al. (2008)	Bulimia Nervosa	52.5	F50.2	Family-Based Treatment	Family Engagement	Disease Symptoms	Estimates of Improvement	36;35	0.2649	0.2384

	Disease	% in age range	ICD	Intervention	Aspect of PFCC	Outcome Category	Outcome	N	g	SEg
le Grange et al. (2007)						Disease Symptoms	Eating Disorder Examination	34;34	0.2651	0.2436
						Disease Symptoms	Beck Depression Inventory	34;34	-0.0880	0.2427
						Disease Symptoms	Rosenberg Self Esteem Scale	34;34	0.1636	0.2429

Note. g = calculated value of Hedges g; SEg = calculated standard error of Hedges g.

Results

Quantitative Meta-Analysis

In general, the 11 identified quantitative papers covered a range of different diseases, intervention types, and outcome measures, with approximately 780 participants investigated across the 11 studies.

A majority of studies and findings (16 findings within 8 of the 11 studies) related to young adults living with psychiatric conditions, particularly schizophrenia and other psychotic disorders, and eating disorders. A large number of interventions were aimed at explicitly including the family in the young person's care (Godart et al., 2012; Grawe, Falloon, Widen, & Skogvoll, 2006; le Grange et al., 2007; McCann et al., 2013; Zaitsoff et al., 2008), or giving the patient and family skills to manage disease outside of the clinic situation (Hagner et al., 2012; Jang, Hwang, Padhye, & Meininger, 2017; Levin & Rotheram-Fuller, 2011; Mackie et al., 2014; Schmidt, Herrmann-Garitz, Bomba, & Thyen, 2016). Few interventions addressed the relationship between patients (and their families) and practitioners, or the introduction of structured goal-setting interventions.

Findings from meta-analysis are presented in forest plots generated using the R metafor package. In each forest plot, study authors are listed in the left-hand column, with the degrees of freedom immediately to the right. Standardised effect sizes and 95% confidence intervals are to the right of each plot, with the calculated random-effects weight in percentage points to the left. A pictorial depiction of the effect size and standard error is in the centre of the plot, with the estimated aggregate effect size in the bottom row of the table in both pictorial form and by numerical value and 95% confidence interval.

Disease Severity.

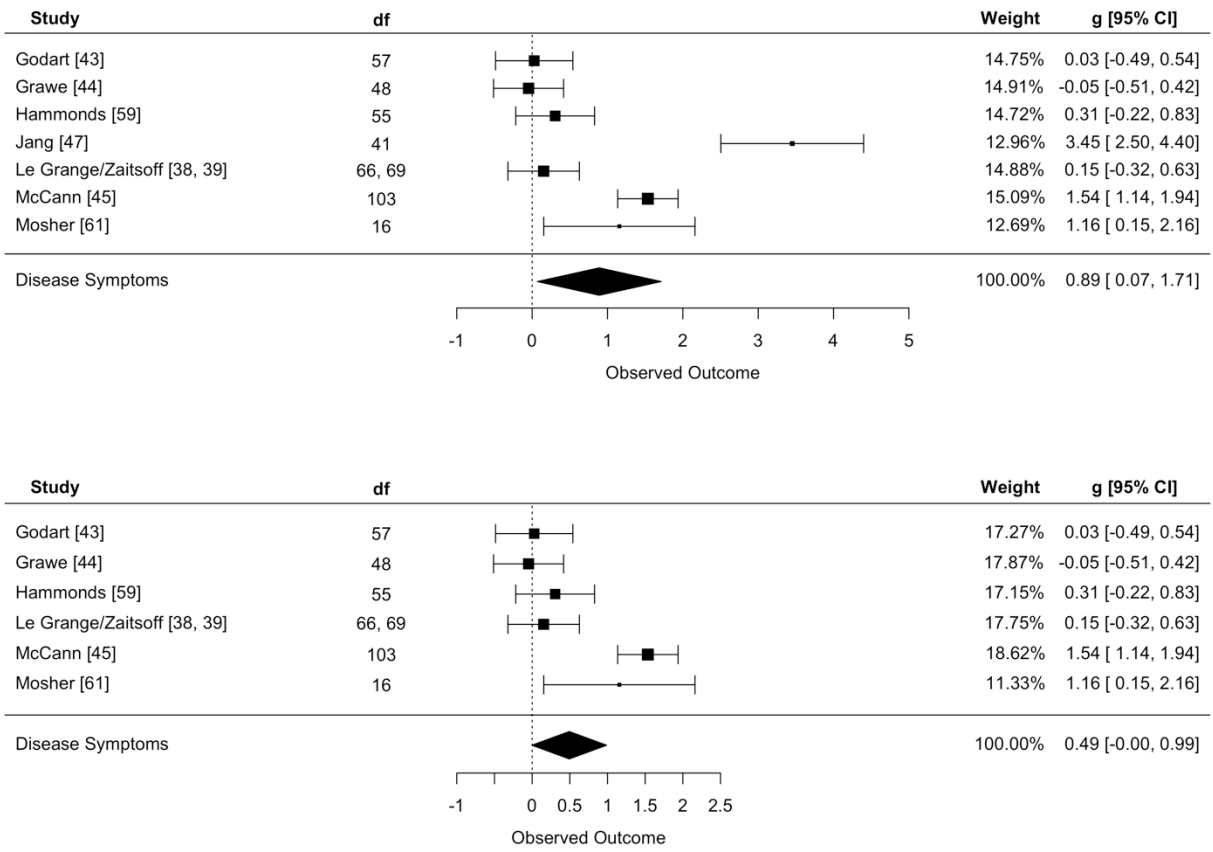


Figure 3.2. Forest plots for Disease Symptoms before and after the removal of a highly influential study. Effect sizes and 95% confidence intervals are shown in the right-most column.

PFCIs were associated with a modest but significant reduction in reported disease severity when all identified papers were included ($g = 0.890$, $p = .034$, see Figure 3.2). However, there was significant heterogeneity and asymmetry in the sample ($I^2 = 94\%$, $z = 2.16$, $p = .030$). In particular, one set of findings exerted significant influence on the analysis (DFBETA = 1.66, Jang et al., 2017). When this was excluded, the combined effect size was positive but not significant ($g = 0.493$, $p = .051$), and significant heterogeneity was still present ($I^2 = 82\%$), although the asymmetry disappeared ($z = 0.50$, $p = .62$) suggesting that any effect on disease severity is modest and highly variable.

Self-Determination/Self-Efficacy.

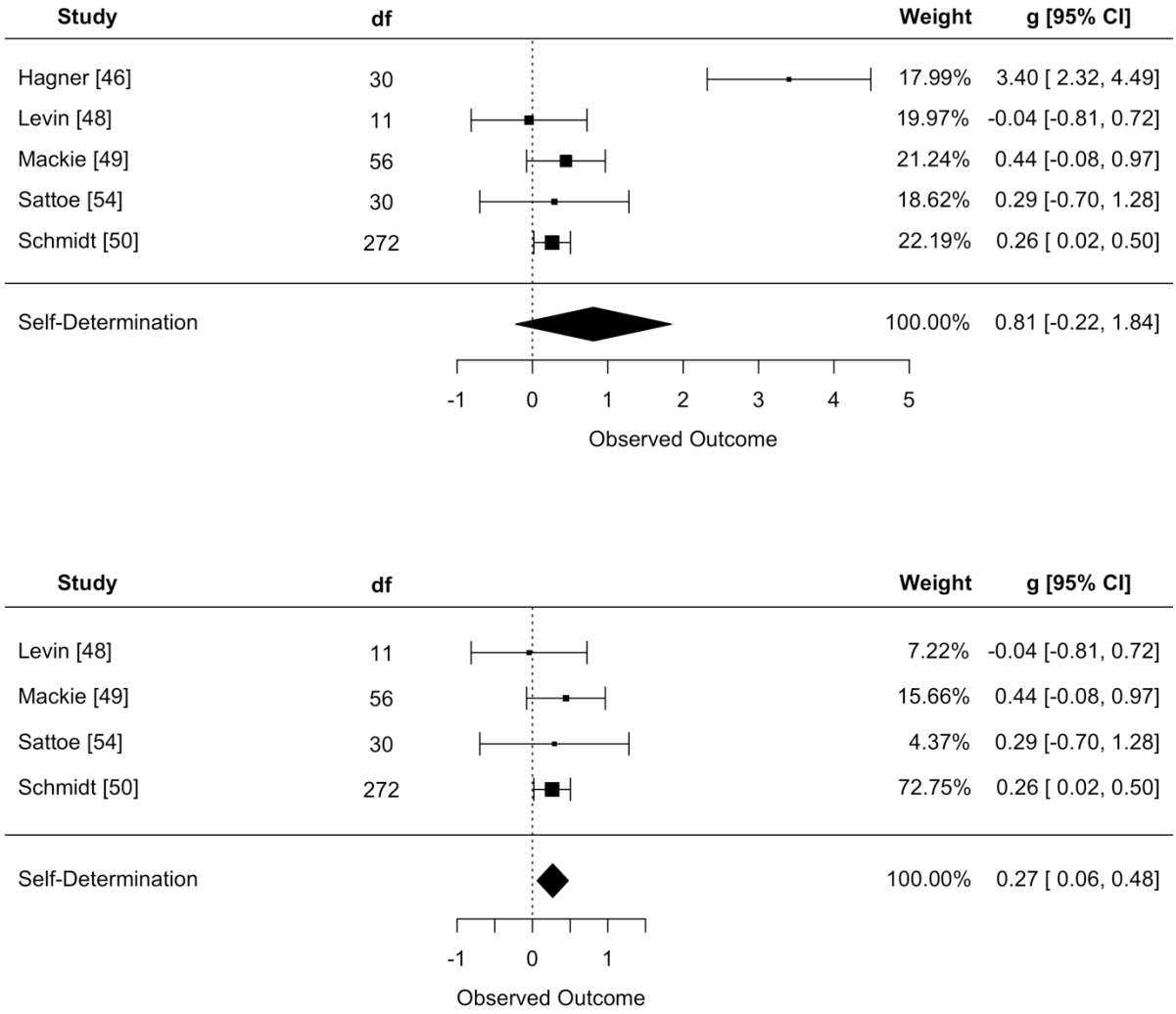
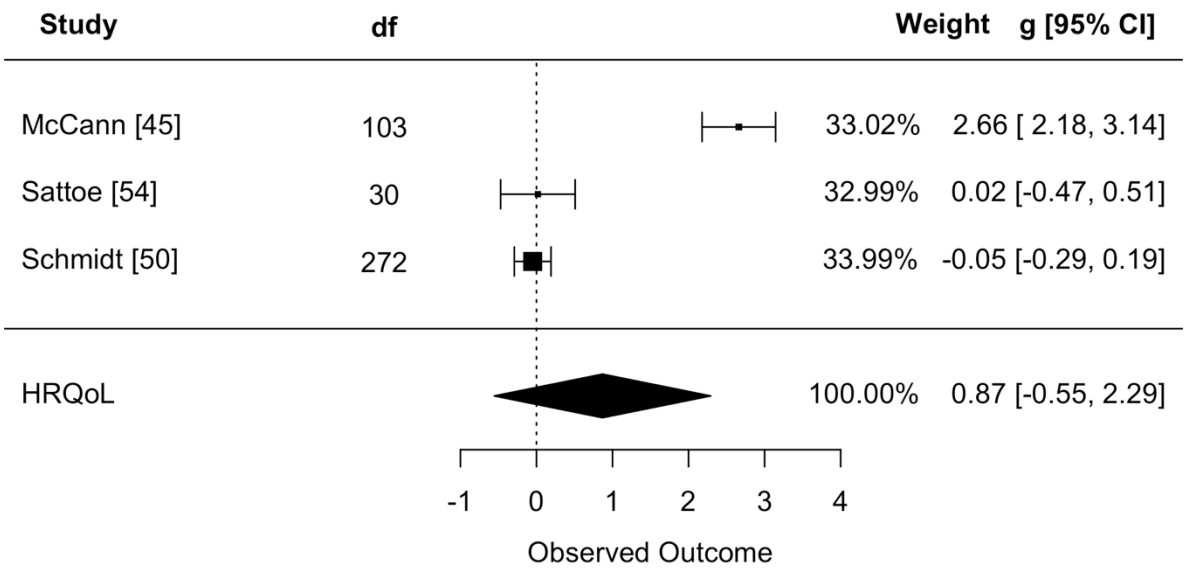


Figure 3.3. Forest plots for Self-Determination/Self-Efficacy status, before and after the removal of a highly influential study.

Effects of PFCIs on self-determination/self-efficacy were moderate and not significantly asymmetrical ($z = 1.54$, $p = .12$). PFCIs tended to produce improvements in self-determination/self-efficacy when all papers were included, although these results were non-significant ($g = 0.810$, $p = .124$, see Figure 3.3). The results were significantly influenced by the inclusion of one paper (DFBETAS = 5.22, Hagner et al., 2012), which studied an intensive intervention compared to no treatment at all. When this paper was excluded, the

aggregate effect, while small, became significantly positive and homogeneous (g = 0.269, p = .011; I^2 = 0%), suggesting that there may be a small but significant effect of PFCIs on the self-determination/self-efficacy status of young people living with chronic health conditions.

HRQoL.



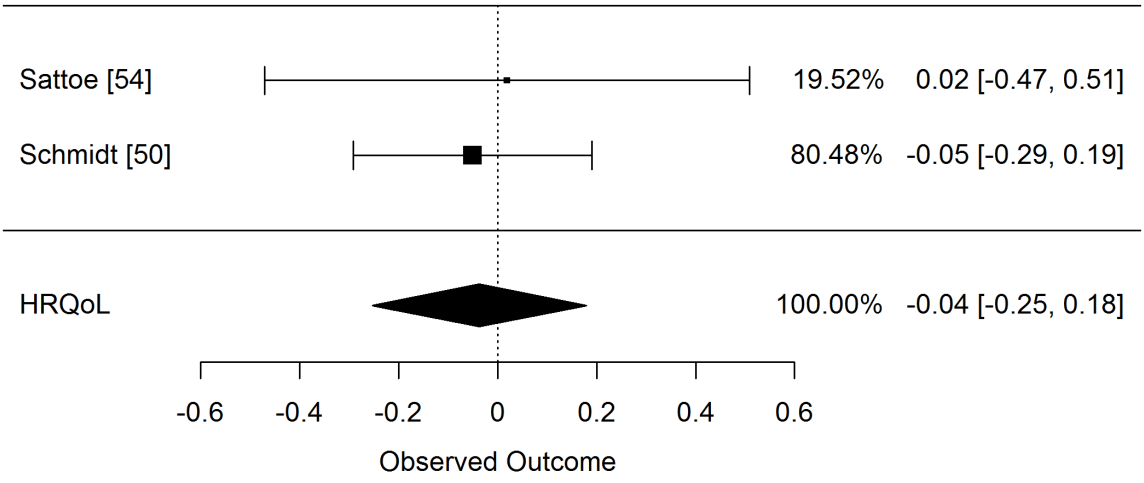


Figure 3.4. Forest plots for Health-Related Quality of Life.

PFCIs tended to produce improvements in HRQoL over standard care, but this effect was non-significant (g = 0.868, p = .232, see Figure 3.4). There was also a large amount of variability in the sample, mainly due to the effect of one finding, which assessed the HRQoL of carers rather than that of patients, although the sample was not significantly asymmetrical (I2 = 97%, z = 1.02, p = .31, McCann et al., 2013). Exclusion of this finding reduced the effect size to effectively zero (g = -0.04, p = .74).

Other measures.

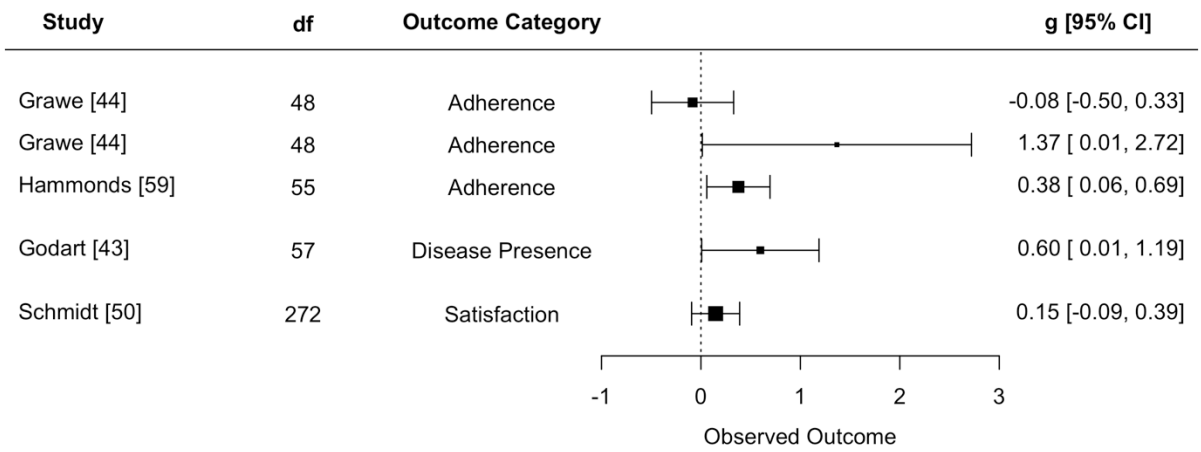


Figure 3.5. Other effects identified in the present review. Note that due to the various kinds of effect reported here, no aggregate effect has been calculated.

In addition to the outcome measures described above, several studies investigated the effects of PFCIs on adherence to treatment, the presence or absence of disease (independent of the severity of disease) and patient satisfaction (Figure 3.5). All effects observed were relatively small, and highly variable. Meta-analysis of these effects was not undertaken due to their different nature, which is an assumption of the technique.

Qualitative Meta-Synthesis

The bulk of studies qualitatively evaluating PFCIs from the perspectives of young people and their families focused on the nature of the interventions and respondents' opinions of and preferences for particular portions of them. However, some information could be extracted addressing patient and family experiences of overall outcome from the identified studies.

In general, qualitative evaluations of PFCIs reported positive effects from the perspective of young people and families. Participants reported that care following a PFCI was "helpful" and "more efficient than expected" (Sawin et al., 2015). In particular, participants felt more confident (Price et al., 2011) and "treated like an adult" where they could assert their independence and make decisions (Price et al., 2011; Sawin et al., 2015) and when they were supported to make plans for the future (Bobier, Dowell, & Craig, 2009). This may reflect the positive effects of PFCIs on self-determination and self-efficacy found in the quantitative literature.

Participants also reported increased empowerment of parents in the health care process (Bobier et al., 2009), which was not addressed in the quantitative studies in the review. Young people also reported that they were "still having problems getting help" (Sawin et al., 2015), suggesting that the implementation of more patient- and family-centred health services does not guarantee high levels of patient satisfaction.

Discussion

The aim of this review of the literature was to determine the impact of interventions to improve the patient- and family-centredness of care (PFCIs) on the effectiveness of care of young adults (16-25) with chronic health conditions. The implementation of PFCIs in the delivery of health services to young adults living with chronic conditions was associated with improvements in the self-determination and self-efficacy of young adult patients, although

the small number of studies available does limit the generalisability of this finding. This work supports the implementation of PFCIs as a means to facilitate young people's engagement with their own health care. Positive trends were also seen for disease severity and HRQoL, although the lack of large, high-quality studies in this area, coupled with significant variability in reported effects, means that these trends were not significantly different from zero.

Very few qualitative studies were identified as part of this study (n=3). Qualitative evaluations of PFCIs seem to support the results of the meta-analysis, in that participants reported higher levels of confidence and increased ability to engage with making decisions. Some studies also found that patients experienced care as more efficient and helpful after implementation of a PFCI, suggesting that these kinds of interventions may have important benefits to how patients experience care as effective, an important component of PFCC (Allen et al., 2018). However, it is difficult to determine whether this is a repeatable effect due to the very small number of qualitative studies.

In addition, two of the three qualitative studies identified as part of this study largely focused on young adult informants, rather than also incorporating the viewpoints of family members, who have previously been identified as important stakeholders in PFCC (Allen et al., 2018). The incorporation of family perspectives is an important part of assessing patient- and family-centred care, as they are likely to experience significant additional burden due to the impact of the chronic health condition, and the study that did incorporate family as informants reported a sense of increased empowerment among parents, which has not been explored in the quantitative literature. As only a single study, however, this result requires significant further investigation, and so future work that explores the impacts of these kinds of interventions on family members is strongly suggested.

Finally, many of the qualitative studies identified in the review focused primarily on the desirability of individual features of the interventions rather than the impacts of the interventions on the experience of care overall, which were secondary findings. This hampers the ability to clearly articulate the effect that PFCIs have on the lived experience of young people and their families. Further investigation into how the experiences of young people living with chronic health conditions and their families are affected by PFCIs is strongly recommended, to explore the effects of PFCIs on the experiences of young people with chronic health conditions and their families.

The studies identified have primarily been performed in the context of mental health disorders. Given the wide range of chronic health conditions affecting young adults, it is not appropriate to draw generalisations from the identified studies to chronic health condition management more broadly. That said, the homogeneity of effect among those studies targeting self-determination/self-efficacy, which included studies targeting young people with renal disease (Schmidt et al., 2016) and diabetes (Sattoe, Jedeloo, & Van Staa, 2013) may suggest that effects are likely to be stable across aetiologies, although this requires additional further investigation.

A large amount of literature reviewed as part of this review contained participants outside the identified participant population, and this is an important limitation of this kind of review. Given the important changes in the lives of young adults over this period, empirical research that focuses on this population in particular is important to determine whether the effects preliminarily identified in this paper are truly present.

While Trim and Fill analysis did not suggest studies with negative findings missing from the sample (an indicator of publication bias), the high variability in effects obtained, coupled with the very small number of studies, may make this determination unreliable. The large number of studies incorporating multiple outcome measures is also a concern, as

collinearity between measures due to patient effects could lead to correlations between the models that violate the assumptions of multiple random-effects modelling. Further high-quality investigations will allow a more stable and comprehensive picture of the impacts of PFCIs on care to emerge, particularly where effects are highly variable such as those observed on disease severity and HRQoL. Additional quantitative investigation into the effects of PFCIs on parent and family member experiences of a young person's care are also warranted, to allow a better understanding of the effects of PFCIs on family trauma due to chronic health conditions.

This review suggests that patient- and family-centred approaches are likely to improve outcomes for patients and families, reflecting similar reviews of PFCC, self-management, and individualised care (Nolte & Osborne, 2013; Qamar, Pappalardo, Arora, & Press, 2011; Radhakrishnan, 2012), although the very small number of studies means that additional research is required. It also reflects the improvement in experiences of treatment and support, and reduced distress during treatment, predicted by the theoretical literature promoting particular approaches to PFCC (Hudon et al., 2012). However, the lack of measurement of outcomes for family members means that this remains an area in which further investigation and research is warranted, to determine what the impacts of chronic disease are on young adults and their families, and how and whether to ameliorate these effects.

This review has also identified that there appears to be a lack of assessment of PFCC in this population that makes use of existing measures of patient-centredness of care, such as the Measure of Processes of Care – Adults (Bamm, Rosenbaum, & Stratford, 2010), the Patient Perception of Patient-Centredness (Hudon et al., 2012), or the Consultation Care Measure(Little et al., 2001). None of these measures have yet been validated for use with young adults living with chronic health conditions, limiting the ability of researchers to

measure patient-centredness and determine its effectiveness. As a result, the validation of these tools in this population, or the development of more appropriate tools to measure and evaluate patient- and family-centredness of care in this population, is of high importance.

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Appendix – Algorithms used to determine fraction of participants who fell within the appropriate age range

Where a mean and standard deviation were reported

```
return NORMDIST(26, mean, sd, TRUE)-NORMDIST(16, mean, sd, TRUE)
```

Where a median and interquartile range were reported

```
return (MIN(26, mdn+1.5*iqr)-MAX(16, mdn-1.5*iqr))/(3*iqr)
```

Where a minimum and maximum were reported

```
return (MIN(26, max)-MAX(16, min))/(max-min)
```

Where multiple bands were reported

```
included, total = 0
for n in n_bands:
     included += (MIN(26, max_n)-MAX(16, min_n))*N_n/(max_n-min_n)
     total += N_n
return included / total
```

CHAPTER 4 – DEMOGRAPHIC AND ACTIVITY STATISTICS OF YOUNG AUSTRALIAN ADULTS LIVING WITH HEARING LOSS: AN OBSERVATIONAL STUDY (SUBMITTED FOR PUBLICATION)

Rationale

Chapters 2 and 3 presented analyses of studies of young adults with a variety of chronic health conditions. However, the ultimate goal of this project was to address the experience of young Australians with hearing loss, with a view towards understanding the aspects of PFCC that apply to them specifically.

However, very little data addressing the life courses, demographic characteristics, and family lives of young adults with hearing losses in Australia previously existed. It was unclear what challenges young adults living with hearing loss experienced, and whether these challenges were more pronounced than those experienced by young adult Australians as a whole. To address this, a study was conducted, in which a variety of demographics were collected from a sample of young adults living with hearing loss, in an attempt to describe their educational attainment, employment situations, histories of audiological (re)habilitation, and happiness with a variety of aspects of life, including overall life satisfaction. These data were compared with existing populations.

It is intended that this paper will be submitted to Trends in Hearing for publication. The results of this study have particular relevance to the delivery of hearing (re)habilitation in the Australian context, several of which are discussed in Chapter 7 of this thesis.

Introduction

Hearing loss is one of the most common disabilities, estimated to affect more than 466 million people around the world (World Health Organization, 2018). Hearing loss affects a large number of young adult (16–25 years old) Australians, with approximately 8000 fitted

with hearing aids or cochlear implants (Australian Hearing, 2018). Hearing loss can have a range of long-term impacts on daily living, including the ability to engage in education, employment, and social relationships (Danermark, Granberg, Kramer, Selb, & Möller, 2013; Vas, Akeroyd, & Hall, 2017).

In children, congenital hearing losses have been shown to be associated with language delay (Vohr et al., 2012), lower self-esteem (Theunissen et al., 2014), and reduced educational outcomes (Davis, Stelmachowicz, Shepard, & Gorga, 1981; National Deaf Children's Society, 2017). The early fitting of hearing devices allows the language outcomes of children with hearing loss to approach those of normally-hearing peers (Ching, Dillon, Leigh, & Cupples, 2018). While it is known that the academic benefits of intensive early intervention can persist beyond secondary schooling (Goldblat & Pinto, 2017), it is unclear whether current audiological (re)habilitation services have benefits in other domains, especially into young adulthood.

Young adulthood, the period of life between the ages of 16 and 25, is a time of significant change across a range of areas of life, marked in part by entry into the workforce (Arnett, 2000). Importantly, employment is a predictor of better mental health (van der Noordt, IJzelenberg, Droomers, & Proper, 2014), and part-time work has been associated with deterioration of physical health compared to full-time work (Ross & Mirowsky, 1995). In addition, difficulty finding employment during young adulthood can lead to significant long-term decrease in earnings (Mroz & Savage, 2006). Hearing disability can be a barrier to finding and maintaining employment (Garramiola-Bilbao & Rodríguez-Álvarez, 2016; Stam, Kostense, Festen, & Kramer, 2013). However, to date, there have been no studies investigating the impact of hearing loss on the ability of young adults to find and maintain employment, a gap which this study aimed to fill.

Young adulthood is also distinguished for many young adults by the development of romantic competence (Davila et al., 2017), demonstrated by their entering into stable romantic relationships and starting families of their own (Arnett, 2000). In Australia, many more 24-year-olds than 18-year-olds are living away from the family home, and are in stable romantic relationships, either de facto or married (National Centre for Vocational Education Research Ltd [NCVER], 2014). Effective interpersonal communication is vital for effective relationships (Knapp & Vangelisti, 2009), and lower communication ability is associated with poorer relationship satisfaction (Meeks, Hendrick, & Hendrick, 1998). Although the social consequences of hearing loss have been studied among older adults (see e.g. Lemke & Scherpiet, 2015), it is unknown whether the communication difficulties caused by hearing loss may lead to reduced satisfaction with romantic and social relationships during young adulthood, with resulting poorer life satisfaction.

Research indicates that adolescents living with disability tend to have poorer life satisfaction when compared with adolescents as a whole (Proctor, Linley, & Maltby, 2009), and this is evident among those who are deaf or hard of hearing (Gilman, Easterbrooks, & Frey, 2004). To date, it is unknown whether such reduced life satisfaction persists beyond secondary schooling and into later life. Adolescents with hearing loss who wear hearing aids have been shown to have lower hearing-related quality of life as compared with adolescents with hearing loss who do not wear hearing aids, potentially due to social stigma associated with hearing aid use (Rachakonda et al., 2013). However, among adults with hearing loss, hearing aids have been reported to improve quality of life (Chisolm et al., 2007). As a result, it is unclear whether hearing aids, hearing (re)habilitation more broadly, or the way in which hearing services are delivered have an impact on the quality of life of young adults with hearing loss.

Patient-centred healthcare, which puts control of the clinical encounter more in the hands of the patient rather than the practitioner (Stewart et al., 2014), is a kind of service delivery that has been associated with a range of positive outcomes, including reductions in mortality (Meterko, Wright, Lin, Lowy, & Cleary, 2010), improved adherence to treatment (Arbuthnott & Sharpe, 2009), and reductions in disability (Diamond, Wenzel, & Nissan, 2006). However, studies with older adults attending audiology services have reported that patient-centred communication is inconsistently implemented (Grenness, Hickson, Laplante-Lévesque, Meyer, & Davidson, 2015a, 2015b). To date, few studies within audiological service delivery have used standardised measures of patient-centredness such as the Measure of Processes of Care – Adults (MPOC-A; Bamm, Rosenbaum, & Stratford, 2010). The measurement of patient-centredness using these kinds of broad and well-established measures has the potential to focus hearing service improvement on those areas of service that are most relevant to patients.

The first aim of this study was to explore the functioning of young adult Australians with hearing loss aged 16-25 years as compared to a national sample of young adult Australians in relation to educational achievement, employment engagement and stability, and happiness with aspects of life including overall life satisfaction. The second aim was to describe the experiences of hearing (re)habilitation for young adults with hearing loss and their use of and satisfaction with hearing devices and services. Finally, this study aimed to determine the association between the experiences of audiological (re)habilitation and overall life satisfaction of young adults with hearing loss.

Method

Study Design

This study used a cross-sectional design, in which an exploratory survey addressed all three research aims in a sample of young adult Australians with hearing loss aged 16-25

years. Australia is unique in that a single, Government-owned hearing provider is funded to provide hearing (re)habilitation services to all people with a confirmed hearing loss under the age of 26, and so the client lists of the government provider were used as the major source of potential participants.

To address the first aim, responses of the sample of young adult Australians with hearing loss to a subset of survey questions were compared to responses to the same questions from a nationally representative sample of young adult Australians who were participants in the Longitudinal Surveys of Australian Youth study (National Centre for Vocational Education Research Ltd [NCVER], 2014). To address the second and third aims, responses of the participants with hearing loss to the remainder of the survey questions were analysed to compare experiences of hearing (re)habilitation, use of and satisfaction with hearing services and devices, patient-centredness of hearing (re)habilitation, and overall life satisfaction with each other.

Materials

The authors developed an online survey (see supplementary material) that included three screening questions, 30 questions selected from the Longitudinal Surveys of Australian Youth (LSAY) study relevant to the research aims, and 18 questions that explored participants' histories and experiences of hearing loss and audiological (re)habilitation. The screening questions ensured that only participants within the appropriate age range, who reported having a hearing loss, and who lived in Australia were included in the sample.

The LSAY is a longitudinal telephone and online survey of a nationally representative sample of young Australians aged between 15-25 years and seeks information about respondents' educational attainment, employment status, and happiness with aspects of life including overall life satisfaction. The questions used in the current study that were adopted from the LSAY survey aimed to elicit demographic information about participants, including

location, gender, language use, educational and employment history, and living situation. Several of these were defined in the LSAY as *Defined Variables*, which are generated from the responses to multiple questions. To enable comparisons with Defined Variables, the research team included questions that would elicit the same data by using the variable descriptions listed in the relevant LSAY Technical Reports (National Centre for Vocational Education Research Ltd, 1995, 2012, 2014, 2015a, 2015b).

In addition, a validated measure of happiness which included overall life satisfaction that was developed for the LSAY was included (question 33; Mahoney, Quested, Thøgersen-Ntoumani, Ntoumanis, & Gucciardi, 2016). This final measure asked participants to rate on a four-point Likert scale (*Very Unhappy*, *Unhappy*, *Happy*, *Very Happy*) how happy there are with a range of aspects of life, including "the work that you do," "your social life," and "your life as a whole" on a four-point Likert scale. In the current study, the item asking "How happy are you with your life as a whole?" was used as a measure of *overall life satisfaction* as suggested by Veenhoven (2012).

Following this set of 30 LSAY questions, a further 18 questions were included in the online survey that elicited general information about: respondents' hearing history (questions 34–37); hearing (re)habilitation history, and use of and satisfaction with hearing devices and services (questions 38–48); and a section asking respondents to rate the importance and delivery of eleven aspects of care identified by the authors as potentially valuable to the patient-centeredness of care in hearing (re)habilitation on a four point Likert scale as part of a previous systematic review (questions 49 and 50; Allen, Scarinci, & Hickson, 2018). The aspects of care presented were: "My audiologist listens to me", "My audiologist explains things clearly", "My audiologist cares about me", "I am involved in making decisions about my hearing loss", "I trust my audiologist to make decisions that are in my best interests", "I can see my audiologist whenever I need to", "I see the same audiologist every time", "My

audiologist addresses my individual needs", "My hearing aids work for me", "I like wearing my hearing aids", and "My audiologist talks to me, not just my parents". Each aspect of care was rated from 1 = *Not Important* to 4 = *Very Important* and from 1 = *Strongly Disagree* to 4 = *Strongly Agree*. The survey then presented the MPOC-A in its entirety (question 51).

The MPOC-A is a measure of the patient-centredness of health care that has been demonstrated to have high internal consistency (alpha from .81 to .93) and test-retest reliability (ICC .73 to .83) in an adult population (Bamm et al., 2010). Respondents in the current study were asked to rate the extent to which their clinicians perform 34 behaviours identified as being patient-centred on a seven-point scale from 1 = *Not at all* to 7 = *To a very great extent*. Participants were also able to mark an item as *Not Applicable*, in which case it was excluded from further calculation. All items were marked as *Not Applicable* by at least two participants, most participants (n = 59) marked at least one item *Not Applicable*, and one participant marked every item *Not Applicable* and was subsequently excluded from analyses of this question. Scores for items that were not marked as *Not Applicable* were then averaged to obtain an overall score (in which all items were included) and five domain scores (in which a subset of items were included) labelled by Bamm et al. as *Enabling and Partnership*, *Providing General Information*, *Providing Specific Information*, *Coordinated and Comprehensive Care*, and *Respectful and Supportive Care*. Higher scores indicate higher levels of patient-centredness. Permission to use the MPOC-A was obtained from CanChild Centre for Childhood Disability Research (www.canchild.ca). In this study, Cronbach's alpha for the domain scores ranged from .89 to .97, and alpha for the overall score was .99.

Participants

Young adults with hearing loss. A total of 2,957 potential young adult participants with hearing loss were identified from the client list of Australian Hearing, a Statutory Authority of the Australian Government that provides hearing services to children,

pensioners, veterans, and other eligible groups. Potential participants were those who were aged between 16 and 26, were recorded as having a hearing loss, and had consented to be contacted for participation in research. Each potential participant was sent a link to the on-line survey. The link was also disseminated via social media, promoted by hearing loss services and advocacy organisations, and by word of mouth. Of those approached for participation, 113 eligible young adults consented to participate and completed the survey, and all were included in analysis.

Young Australian adults. Respondents to the initial five waves of the LSAY (those recruited at 15 years old in 1995, 1998, 2003, 2006, and 2009) were included in the current study (a total of 66,521 participants). LSAY participants were resurveyed annually for 10 years. Responses collected in years up to and including 2016 were included, leading to 373,999 completed surveys (Australian Department of Education and Training, 2017a, 2017b; Australian Department of Education Employment and Workplace Relations, 2017a, 2017b; Australian Government Department of Education, 2017).

Demographic details of the two participant samples are reported in Table 4.1. Young adults with hearing loss who completed the online survey were not significantly different in age to the sample of LSAY observations ($t = –0.3$, $p = .75$). There was a significantly higher proportion of female respondents with hearing loss to the online survey compared to both respondents to the LSAY ($z = –3.5$, $p < .001$) and people aged 0 to 25 fitted with hearing devices in Australia (48.5%, $n = 24544$, $z = –3.2$, $p = .002$; Australian Hearing, 2018). Young adults with hearing loss in the present study were slightly more likely than young adult Australians as a whole to use a language other than English around the home, and less likely to report being of Aboriginal or Torres Strait Islander background (2% in the present study, 6% in the LSAY and Australian Hearing data).

Table 4.2 shows hearing-related demographic details of young adults with hearing loss recruited as part of the present study, and relevant comparison data for young adults (aged 16 to 25) fitted with hearing devices in Australia (as reported by Australian Hearing, 2018). The 113 respondents to the online survey reported significantly greater degrees of hearing loss than young adults fitted with hearing devices in Australia, with 85% reporting a hearing loss of moderate or worse, compared to 37% as reported by Australian Hearing as of the end of 2017 ($U(109, 25155) = 2000000$, $p < .001$, 2018). Reported hearing device usage in the present data appears similar to that previously reported in adult hearing aid users, with around the same proportion of young adults in the current study reporting that they used their hearing device more than 8 hours per day (55%; Hickson, Clutterbuck, & Khan, 2010).

Table 4.1

General demographic details of young adult respondents with hearing loss who completed the online survey in the present study, LSAY respondents, and Australians aged 16 to 25 fitted with hearing aids by the Government-funded hearing services provider

	Young Adults with Hearing Loss (n = 113)	**LSAY (n = 66,521) (n_{obs} = 373,999)**
Age (yrs)	20 (2.80)	19 (3.1)*
Gender (Female)	75 (66%)	33,302 (50%)
Language use		
- Primarily English	101 (89%)	58,769 (91%)
- English and spoken language other than English	5 (4%)	Not Collected
- English and Auslan	5 (4%)	Not Collected
- Language other than English	2 (2%)	5,622 (9%)
Aboriginal or Torres Strait Islander Identity	2 (2%)	3,639 (6%)
Marital Status		
- Single	101 (89%)	164,661 (83%)*

	Young Adults with Hearing Loss (n = 113)	LSAY (n = 66,521) (n_{obs} = 373,999)
- De Facto	11 (10%)	25,074 (13%)*
- Married	1 (1%)	7,476 (4%)*
- Separated	0	342 (0%)*
- Divorced	0	63 (0%)*
- Widowed	0	22 (0%)*
Hours worked per week (hrs)	22.0 (16.2)	25.0 (16.0)*
Weekly take-home pay (AUD)	451 (408)	418 (469)*
Employed		
- Unemployed	47 (42%)	127,163 (35%)*
- Full-time	22 (19%)	94,004 (26%)*
- Part-time	44 (39%)	145,558 (40%)*
Employment Stability		
- Permanent	38 (58%)	96,140 (46%)*
- Casual	28 (42%)	111,437 (54%)*
Unemployed during the previous year	45 (40%)	66,927 (22%)*
Same employer as the previous year	75 (66%)	109,808 (49%)*

* = Calculated per-observation, not per-person

Table 4.2

Self-reported hearing-related demographics of young adult respondents with hearing loss (n = 113) and comparison demographics of Australians aged from 16 to 25 fitted with hearing aids by the Government-funded hearing services provider (n = 7949)

Age at Diagnosis	At birth	< 5 years	Primary School	High School	After High School	Don't Know
- Survey sample (self-report)	**17 (15%)**	**48 (42%)**	**18 (16%)**	**18 (16%)**	**11 (10%)**	**1 (1%)**
- Australian Hearing (2018)	**239 (3%)**	**2354 (30%)**	**2714 (34%)**	**1666 (21%)**	**976 (12%)**	
Hearing Loss Laterality	Bilateral	Unilateral				
	85 (75%)	**28 (25%)**				
Hearing Loss Degree	Mild	Moderate	Severe	Profound	Total	Don't Know
- Survey sample (self-report)	**17 (15%)**	**46 (41%)**	**24 (21%)**	**17 (15%)**	**5 (4%)**	**5 (4%)**
- Australian Hearing (2018)	**4273 (54%)**	**1758 (22%)**	**1046 (13%)**	**866 (11%)**		**6 (0%)**
Hearing Loss Type	Sensorineural	Conductive	Mixed	Central	Don't Know	
	32 (28%)	**9 (8%)**	**12 (11%)**	**1 (1%)**	**59 (52%)**	
Use of hearing devices	8 hrs/day	4–8 hrs/day	1–4 hrs/day	Occasionally (1 hr/month)	Rarely	Never
	62 (55%)	**19 (17%)**	**5 (4%)**	**11 (10%)**	**9 (8%)**	**7 (6%)**
Satisfaction	Very Satisfied	Satisfied	Unsatisfied	Very Unsatisfied		
Hearing Devices	**47 (42%)**	**47 (42%)**	**10 (9%)**	**7 (6%)**		
Hearing Services	**71 (63%)**	**29 (26%)**	**9 (8%)**	**4 (4%)**		
Duration of:	> 10 years	5–10 years	1–5 years	< 1 year	Don't Know	

Attending hearing services	**77 (68%)**	**5 (4%)**	**24 (21%)**	**7 (6%)**	**0 (0%)**
Regular audiologist (n = 71)	**9 (8%)**	**11 (10%)**	**35 (31%)**	**14 (12%)**	**1 (1%)**
When attending the audiologist:	Mother/Stepmother	Father/Stepfather	Sister	Non-relative	Intimate Partner
Who do you take? (n = 60)	**56 (93%)**	**17 (28%)**	**3 (5%)**	**2 (3%)**	**3 (5%)**
Who would you like to take? (n = 53)	**33 (62%)**	**6 (11%)**	**3 (6%)**	**7 (13%)**	**12 (22%)**

Procedure

Access to LSAY raw datasets was approved by the National Centre for Vocational Education Research and the Australian Data Archive. The survey research with young adults was approved by The University of Queensland Human Research Ethics Committee, the Australian Hearing Ethics Committee, and the Hear and Say Ethics Committee.

Upon visiting the survey web page, reading information about the study, and being deemed appropriate for inclusion, participants in the sample of young adults with hearing loss were asked to complete the survey in its entirety. Proceeding to the completion of the study was considered to be informed consent. After the data collection period of 34 weeks concluded, the online survey was closed, and responses were downloaded and analysed.

Statistical Analysis

Analyses were conducted using SciPy 1.0.1 (Jones, Oliphant, & Peterson, 2001), Pandas 0.22.0 (McKinney, 2011), statsmodels 0.8.0 (Seabold & Perktold, 2010), and IPython 6.3.1 (Perez & Granger, 2007). Due to the large number of comparisons made, $\alpha = .025$ was used for determining statistical significance (Benjamini & Hochberg, 1995).

LSAY data were weighted using provided LSAY "sample and attrition" weightings, which corrected variability in sampling and non-random loss to follow-up to create a more representative sample. These weightings combine a "sample" weighting that takes into account differences in location and school of initially sampled participants to create an initial weighted sample that is representative of 15-year-old Australians, and an "attrition" weighting that accounts for non-uniform attrition from year to year in such a way that weighted samples at each age are representative of the demographics of the initial participants (Lim, 2011; Rothman, 2009).Weightings were renormalised within each year of collection to sum to the total number of participants in each wave to remove the effect of differing sample sizes between waves.

The effect of hearing loss on continuous variables (e.g., weekly take-home pay, hours worked) was estimated using least squares regression, and the effect of hearing loss on binary variables (reported as an odds ratio, denoted e^B) was estimated using logistic regression. Where nonparametric tests (Spearman rho and Mann-Whitney U) were used, weightings were not used.

Due to the strong dependence of variables such as level of education, living at home status, and marital status on age and gender, the variables age and gender were controlled for in all parametric analyses.

Results

Comparison of Young Adults with Hearing Loss to Respondents to the LSAY

Characteristics describing education, employment and happiness of the 113 young adults with hearing loss were compared to those of participants of the LSAY. In regard to educational achievement, young adults with hearing loss were not significantly less likely than the LSAY sample to complete Year 12, the final year of post-compulsory secondary education in Australia ($e^B = 1.6$, $p = .17$). Among respondents who completed Year 12, young adults with hearing loss were also not significantly less likely to continue to a Bachelor Degree ($e^B = 1.6$, $p = .027$).

From an employment perspective, young adults with hearing loss who responded to the survey reported significantly fewer weekly hours worked ($t = -2.4$, $p = .018$), but did not report significantly different mean total weekly take-home pay ($t = -1.1$, $p = .28$) compared to LSAY respondents. Respondents with hearing loss were also significantly less likely to be currently employed ($e^B = 0.45$, $p < .001$), and more likely to have been unemployed during the previous year ($e^B = 2.3$, $p < .001$). Young adults with hearing loss who were working were significantly less likely to be working full-time than young Australians as a whole ($e^B = 0.45$, $p = .0049$). Interestingly, however, they were significantly more likely to have retained

the same employer for the previous 12 months ($e^B = 2.2$, $p < .001$), and were not significantly less likely to be employed on a permanent basis ($e^B = 1.3$, $p = .38$), suggesting increased employment stability over time.

Table 4.3 shows a comparison of the distributions of happiness within each aspect of life between young adults with hearing loss who completed the online survey, and respondents to the LSAY. Young adults with hearing loss reported lower happiness in all aspects of life than respondents to the LSAY, and significantly lower happiness in a majority of aspects of life.

Table 4.3

Happiness of young adult respondents with hearing loss (n = 113) compared with LSAY respondents using Mann-Whitney U.

How happy are you with…	**U (× 10^6)**	**p**	**n_{LSAY}**
Your future?	12	<.001	267,879
The work that you do?	13	<.001	270,503
What you do in your spare time?	13	<.001	271,701
How you get on with people?	11	<.001	271,931
The money you get each week?	11	<.001	270,720
Your social life?	10	<.001	271,775
Your independence?	13	<.001	271,752
Your career prospects?	13	<.001	268,858
Your standard of living?	14	.08	271,912
The way the country is run?	10	.28	186,916
The state of the economy?	9	.0055	179,613
Where you live?	15	.17	271,777
Your life at home?	14	.12	271,738
Your life as a whole?	11	<.001	271,650

The Effectiveness and Patient-Centredness of Hearing (Re)habilitation

The 113 young adults with hearing loss in the current study reported high satisfaction with hearing services and hearing devices, with the majority of participants reporting that they were *Satisfied* or *Very Satisfied* on both questions (see Table 4.2). Hearing service satisfaction was significantly higher among those who had a regular audiologist than among those who did not ($U = 1943$, $p < .001$), although this was not associated with increased length of relationship with a regular audiologist ($r_s = .22$, $p = .059$) or severity of hearing loss ($r_s = .098$, $p = .31$). Higher hearing device satisfaction was significantly associated with increased frequency of hearing device use ($r_s = .42$, $p < .001$), but was not significantly associated with having a regular audiologist ($U = 1500$, $p = .48$), the length of relationship with a regular audiologist ($r_s = .21$, $p = .089$), or the severity of hearing loss ($r_s = -.13$, $p = .19$). Reported severity of hearing loss was not associated with whether a person reported wearing hearing devices or not ($U = 281.5$, $p = .13$) or how often respondents who wore hearing devices used them ($r_s = .07$, $p = .45$). Finally, there was no difference in the frequency of device use between those respondents living at home and those who did not live at home ($U = 905$, $p = .16$).

Patient-centredness in this population was assessed in two ways: (1) by participants rating eleven aspects of patient-centred care identified in a previous systematic review by the authors as potentially valuable to the patient-centeredness of care in hearing (re)habilitation on both their importance to the participant and whether the participant perceived that they were experiencing them, and (2) using the MPOC-A. All aspects of patient-centred care identified by the authors were rated as either important or very important by the majority of respondents (question 49), with the majority of respondents agreeing or strongly agreeing that each was being delivered within audiological (re)habilitation (question 50). There was a significant positive correlation between importance of a patient-centredness behaviour and

delivery of this behaviour by audiologists as reported by respondents (r_s = .49, p < .001), indicating that aspects of care that participants described as more important were also those which they more strongly agreed that they experienced. Due to strong ceiling effects, there was insufficient variability in these ratings to facilitate meaningful additional analysis, and they are therefore unlikely to be useful in the future as a measure of patient-centredness or desirability of audiological (re)habilitation.

Table 4.4

Ratings of patient-centredness of audiology care by young adult respondents with hearing loss as measured by the MPOC-A (n = 113)

Domain	**Mean Score**	**SD**	**Range**	**α**
Overall	23.8	8.17	[0–30]	.988
Enabling and Partnership	4.96	1.41	[0–6]	.968
Providing General Information	4.40	1.80	[0–6]	.901
Providing Specific Information	4.31	1.94	[0–6]	.898
Coordinated and Comprehensive Care	4.78	1.61	[0–6]	.946
Respectful and Supportive Care	5.09	1.40	[0–6]	.960

MPOC-A scores both overall and within each of the five domains were generally high, indicating high patient-centredness of care delivered by audiologists, and are listed, along with Cronbach's alphas, in Table 4.4. There was a large variability in MPOC-A scores, with the maximum possible range (0–30 out of 30). Overall MPOC-A score was not associated with participants' reports of the severity of their hearing loss (r_s = −.006, p = .50), duration of hearing loss (r_s = .12, p = .20), or duration of regularly seeing an audiologist (r_s = −.12, p = .19). MPOC-A scores were significantly higher among respondents who reported having a regular audiologist (U = 1103, p = .016).

The Association Between Experiences of Audiological (Re)habilitation and Overall Life Satisfaction

There was a significant positive correlation between overall MPOC-A score and overall life satisfaction as measured by the item "how happy are you with your life as a whole?" (r_s = .270, p = .004), although there was no difference in overall life satisfaction between those who had a regular audiologist and those who did not (U = 1458, p = .42). Overall life satisfaction was not associated with hearing loss severity (r_s = −.01, p = .87), satisfaction with hearing services (r_s = .13, p = .17), satisfaction with hearing devices (r_s = .00, p = .998), frequency of device use (r_s = .05, p = .62), or length of relationship with a regular audiologist (r_s = −.02, p = .90).

Discussion

The aims of this study were: (1) to explore the functioning of young adult Australians with hearing loss aged 16-25 years as compared to a national sample of young adult Australians in relation to educational achievement, employment engagement and stability, and happiness with aspects of life including overall life satisfaction; (2) to describe the experiences of hearing (re)habilitation for young adults with hearing loss and their use of and satisfaction with hearing devices and services; and (3)to determine the association between the experiences of audiological (re)habilitation and overall life satisfaction of young adults with hearing loss

In general, young adults with hearing loss reported similar demographics and educational and employment situations to young adult Australians as a whole, indicating that employment and education demographics of young Australians at large are likely to translate well to young adults with hearing loss, and so may be useful for large-scale planning of hearing services for young adults. However, recognising the particular challenges faced by young adults with hearing loss is also important for hearing services delivery. Overall, most

respondents reported that they were happy with the clinical services they were receiving, although the large variability across the respondents means that there is still opportunity to further improve satisfaction with audiological (re)habilitation for some young adult patients.

The high levels of educational engagement among young people with hearing loss who responded to this study are encouraging. Participation in secondary education beyond Year 10 is not compulsory in Australia, and Signing Deaf Australians have previously reported lower levels of Year 12 completion than their hearing peers (ORIMA Research, 2004, p. 6), although no difference was found for the sample in the present study, suggesting that non-signing young people with hearing loss may not be subject to this same trend. This extra time in schooling represents an important opportunity for interventions to ensure that young people with hearing loss are prepared for the transition to university or into the workforce. While it is known that young people with hearing loss require longer to achieve successful transition out of secondary schooling, little is known about the services that are available to support this transition, and so further research is recommended (Luft, 2014). That many young people with hearing loss continued on to tertiary study is also encouraging, as it suggests that young people with hearing loss are not being dissuaded from non-compulsory education by their hearing losses. In Australia, many tertiary educational institutions access Government funding to provide educational supports for young people with disabilities including hearing loss (KPMG, 2015). Further research to understand the needs of young adults with hearing loss may assist tertiary institutions to provide more targeted support to students in addition to those that may already be available, such as notetaking, transcribed lectures, and wireless communication systems.

This additional opportunity for intervention is particularly important when considered in the light of findings in this study of higher unemployment rates and lower hours of employment among the sample of young adults with hearing loss compared to a population

sample of young adults in Australia. While young people with hearing losses who achieved employment were not significantly disadvantaged financially compared to young Australians as a whole (and in fact were more likely to have stability in their employment), increasing rates of unemployment and underemployment among young adults in Australia may have a disproportionate impact on those living with hearing loss (Australian Bureau of Statistics, 2018; Brotherhood of St Laurence, 2017). The poorer happiness with regards to employment and career prospects expressed by young people with hearing loss further underlines the importance of support services and tailored audiological (re)habilitation to assist this population to gain and maintain employment.

Young adults with hearing loss reported significantly reduced happiness with "get[ting] on with people" and "social life" compared to young Australian adults as a whole. This may indicate that they are experiencing increased communication difficulties in more complex social situations with high levels of background noise. Social interactions often feature more than one talker and extraneous noise sources, which are more difficult for people with hearing loss to filter out than their normally-hearing peers (Dai, Best, & Shinn-Cunningham, 2018), and difficulty in social environments remains an important source of disability for people with hearing loss (Danermark et al., 2013). As a result, interventions targeted at multi-talker social situations may be particularly effective in this population. For example, communication education that develops problem solving skills to improve functioning in challenging environments has been shown to be effective in older adults (e.g. Hickson, Worrall, & Scarinci, 2007), but these kinds of programmes have not been tested for young adults with hearing loss.

The association between higher levels of reported patient-centredness of care and overall life satisfaction found in this study underscores the potential importance of patient-centred care. While having a regular audiologist improves the delivery of patient-centred care

as defined by the MPOC-A, the lack of association between having a regular audiologist and overall life satisfaction suggests that it is the patient-centred care itself that improves overall life satisfaction. As a result, targeting continuity of care as an outcome is not sufficient to guarantee positive overall life satisfaction outcomes for these patients. The unique nature of current hearing service delivery in Australia, in which a young person is only able to access their Government-funded audiologist until they turn 26, may threaten this continuity of care further, although upcoming changes to service delivery may enable young people to continue seeing their regular audiologist beyond this point.

Respondents in the present study report much higher levels of patient-centredness of care than in the only other study that could be located applying the MPOC-A in audiological (re)habilitation situations (Ali, 2018). This is heartening, as it suggests that audiologists in Australia are generally reported by young adults with hearing loss to be meeting their needs in a strongly patient-centred way. However, the wide ranges of responses from participants suggest that some young adult patients are not receiving these kinds of care, which represents an opportunity for audiologists to increase the patient-centredness of hearing (re)habilitation practice. The application of existing models of patient-centred care that prioritise the patient's experience, consistency of care, and shared planning and decision-making should be prioritised for this patient group (e.g. Grenness, Hickson, Laplante-Lévesque, & Davidson, 2014; Laplante-Lévesque, Hickson, & Worrall, 2010; Stewart et al., 2014).

A proportion of young people in the current study reported never or rarely wearing hearing devices (14%), and this did not decrease with increased self-reported severity of hearing loss. Poor adherence to treatment among adolescents and young adults has been noted across a range of chronic health conditions (Bishop, Lemberg, & Day, 2014; Borus & Laffel, 2010; Denison et al., 2015; Kondryn, Edmondson, Hill, & Eden, 2011), with hearing loss no exception. Application of patient-centred care has been shown to improve adherence

to treatment in other chronic health conditions (Arbuthnott & Sharpe, 2009; Kondryn et al., 2011) and so the application of these kinds of models may help improve hearing device use for those who are choosing not to wear hearing aids at all. From diagnosis in infancy, hearing aid usage is driven largely by parental involvement (McCracken, Young, & Tattersall, 2008), and parents play an important role in maintaining audiological (re)habilitation during childhood: as young people establish independence from family, one of the areas in which they can exercise their sense of autonomy may be the decision to wear or not to wear hearing devices. Importantly, the presence of family members in the home is not sufficient to explain this disparity, and so interventions to address the needs of young people who choose not to wear hearing devices need to begin well before they move away from the family home, similar to transition interventions for young people living with other chronic health conditions (Fegran, Hall, Uhrenfeldt, Aagaard, & Ludvigsen, 2014).

Young adults with hearing loss who responded to the online survey reported a variety of family members who might be considered relevant from the perspective of clinicians for inclusion in hearing (re)habilitation, described in Table 4.2. A majority of respondents identified their parents, particularly mothers/stepmothers, as being the people who they are both most likely to actually bring or wish to bring with them to audiological appointments. The inclusion of parents is strongly recommended as a part of patient- and family-centred care (Committee On Hospital Care & Institute For Patient- and Family-Centered Care, 2012). When treating young adults, maintaining the patients' independence and autonomy requires that parents be a partner in the health service delivery process while the patient is the primary focus of information and attention (Allen et al., 2018), and so the creation of clinical situations in which parents are able and welcomed to attend is important even as young adults become older.

Demographic details of the young adults with hearing loss in the present study highlighted that 12% use a language other than English as a primary language around the home, which may indicate that communication using interpreters or other language supports may be more effective, comfortable, and empowering for them. Access to interpreters is limited in Australia, with a large number of people relying on family or friends to interpret for them (ORIMA Research, 2004, p. 12). While it may be tempting to rely solely on English to communicate, or to use family members to interpret, the use of family as interpreters is not considered best practice as it runs the risk of omissions or inaccurate translation (Hilder et al., 2017).

This study has several important limitations, primarily due to the relatively small sample compared to the population size. Of particular concern is the apparent gender bias in the sampling. Best available data suggests that young people with hearing loss are approximately as likely to be male as to be female (Australian Hearing, 2018). As a result, this survey has significant gender-based response bias, not dissimilar to existing research on online survey response rates (Sax, Gilmartin, & Bryant, 2003). The lower response rate of young male adults to engage with online survey research may suggest that other methods of data collection are more useful when targeting young adults with hearing loss, potentially by utilising their existing health appointments.

The over-representation of moderate to severe hearing losses in the sample compared to available data for young people fitted with hearing devices in Australia may suggest either a response bias towards young people with greater hearing losses, or an overestimation of the level of hearing loss experienced by young people. In previous studies of self-reported hearing loss, respondents with hearing loss tend to under-estimate their hearing losses compared to behavioural testing (Sindhusake et al., 2001), although it is known that the prevalence of moderate hearing losses tends to be greater for young adults than adolescents

among those fitted with hearing devices in Australia (Australian Hearing, 2018, p. 6). The lack of an association between reported severity of hearing loss and satisfaction with hearing services, satisfaction with hearing devices, or hearing device use does suggest that this overrepresentation is unlikely to be biasing the results in a great fashion, although more work targeting those young adults with milder hearing losses who make up the bulk of young people living with hearing loss in these populations may be warranted.

Conclusions

On the whole, this study suggests that young adult Australians living with hearing loss are not remarkably different to young adult Australians at large, although hearing loss does appear to present additional challenges to education, employment, and overall life satisfaction. Hearing services are experienced positively by young adult patients, although work still needs to be done to deliver high-quality audiological (re)habilitation that meets the needs of all young adult Australians living with hearing loss. Through increasingly patient-centred care among this population, it is possible that young adults living with hearing loss may achieve life satisfaction results similar to those of their normally-hearing peers.

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Supplementary Materials – Survey instrument

This section replicates the survey instrument presented to respondents, along with the corresponding variables in the LSAY data set where these comparisons were made. Questions that have been excluded from analysis due to low response numbers are marked with an asterisk (*).

Question Number	Question Text	Response Options	Corresponding Variable in LSAY Data Set
1	How old are you?	_____	age
2	Do you currently have a hearing loss?	☐ Yes ☐ No	N/A
3	What country do you currently live in?	_____	N/A
4	What is your postcode?	_____	PC
5	What is your gender?	☐ Male ☐ Female ☐ Other ☐ Prefer not to say	Various
6	Are you an Aboriginal or Torres Strait Islander person?	☐ Yes, Aboriginal ☐ Yes, Torres Strait Islander ☐ Yes, both ☐ No	INDIG
7	What languages do you use at home? e.g. English, Auslan, Signed	_____	Various

	English, Mandarin You may enter more than one		
8	What is the language you prefer to use?	_____	Various
9	Are you currently studying at secondary school?	☐ Yes ☐ No → go to 11	XCSL
10	What year are you in?	☐ Year 9 ☐ Year 10 ☐ Year 11 ☐ Year 12	XCSL
11	What is the highest level of school you have completed?	☐ Year 9 ☐ Year 10 ☐ Year 11 ☐ Year 12	XHSL
12	Are you currently studying a post-secondary qualification?	☐ Yes ☐ No → go to 15	XCEL
13	What level qualification is it?	☐ Certificate I ☐ Certificate II ☐ Certificate III ☐ Certificate IV ☐ Diploma ☐ Advanced Diploma	XCEL

		☐ Bachelor Degree ☐ Graduate Certificate ☐ Graduate Diploma ☐ Postgraduate Degree	
14*	Are you studying full-time or part-time?	☐ Full-time ☐ Part-time	XFTS
15	What is the highest level of post-secondary qualification that you have completed?	☐ Certificate I ☐ Certificate II ☐ Certificate III ☐ Certificate IV ☐ Diploma ☐ Advanced Diploma ☐ Bachelor Degree ☐ Graduate Certificate ☐ Graduate Diploma ☐ Postgraduate Degree	XHEL
16	Have you ever enrolled in a Bachelor's level qualification?	☐ Currently undertaking ☐ Completed ☐ Completed and undertaking further study ☐ Commenced, but did not complete ☐ Never commenced	XBAC
17*	Have you ever enrolled in a VET qualification?	☐ Currently undertaking ☐ Completed ☐ Completed and undertaking further study	XVET

		☐ Commenced, but did not complete ☐ Never commenced	
18*	Have you ever commenced an apprenticeship or traineeship?	☐ Currently undertaking ☐ Completed ☐ Completed and undertaking further study ☐ Commenced, but did not complete ☐ Never commenced	XATR
19	Are you currently employed?	☐ Yes ☐ No → go to 24	XLFS
20	How many hours do you currently work a week, on average?	_____	XHRS
21	Are you currently working full-time or part-time?	☐ Full-time ☐ Part-time	XFTP
22	Are you employed on a permanent or a casual basis?	☐ Permanent ☐ Casual	XEMP
23	What is your weekly take-home pay?	_____	XWKP
24	Have you been unemployed during the last year?	☐ Yes ☐ No → go to 26	XUNE
25*	In the last year, how many weeks have you been unemployed for?	_____	
26	Have you changed jobs in the last year?	☐ No – same employer as a year ago ☐ Yes – different employer to a year ago	XMOB

		☐ Yes – gained employment this year ☐ Yes – lost employment this year	
27	What is your marital status?	☐ Married ☐ De facto ☐ Single ☐ Separated ☐ Divorced ☐ Widowed	XMAR
28	What is your living situation?	☐ Living with family ☐ Own outright/buying ☐ Renting ☐ Other	XATH, XOWN
29	How many people currently live with you? Do not include yourself.	_____	Various
30	What relationship do you have to the people living with you?	☐ Father/stepfather ☐ Father-in-law/Partner's father ☐ Mother/Stepmother ☐ Mother-in-law/Partner's Mother ☐ Spouse/De facto ☐ Partner ☐ Brother ☐ Sister	Various

		☐ Boyfriend/Girlfriend ☐ Own children/Stepchildren ☐ Other relatives ☐ Non-relatives				
31*	Do you have any dependent children?	☐ Yes ☐ No → go to 33				XCHI
32*	How many children are dependent on you?	_____				XCHI
33	How happy are you with:	Very Happy			Very unhappy	Various
	Your future	☐	☐	☐	☐	
	The work that you do	☐	☐	☐	☐	
	What you do in your spare time	☐	☐	☐	☐	
	How you get on with people	☐	☐	☐	☐	
	The money you get each week	☐	☐	☐	☐	
	Your social life	☐	☐	☐	☐	
	Your independence	☐	☐	☐	☐	
	Your career prospects	☐	☐	☐	☐	
	Your standard of living	☐	☐	☐	☐	
	The way the country is run	☐	☐	☐	☐	
	The state of the economy	☐	☐	☐	☐	
	Where you live	☐	☐	☐	☐	

	Your life at home	☐	☐	☐	☐	
	Your life as a whole	☐	☐	☐	☐	
34	When were you diagnosed with a hearing loss?	☐ At birth ☐ 0–5 years ☐ Primary school ☐ High School ☐ After High School ☐ I don't know				
35	Is your hearing loss in one ear or both?	☐ Unilateral (in one ear only) ☐ Bilateral (in both ears) ☐ I don’t know				
36	What kind of hearing loss do you have?	☐ Sensorineural ☐ Conductive ☐ Central ☐ Mixed ☐ I don’t know				
37	How severe is your hearing loss?	☐ Mild ☐ Moderate ☐ Severe ☐ Profound ☐ Total ☐ I don’t know				

38	How long have you been going to audiologists?	☐ Less than a year ☐ 1–5 years ☐ 5–10 years ☐ > 10 years ☐ I don't know	
39	Do you have a regular audiologist who you see for most of your appointments?	☐ Yes ☐ No → go to 41	
40	How long have you been seeing that audiologist?	☐ Less than a year ☐ 1–5 years ☐ 5–10 years ☐ > 10 years ☐ I don't know	
41	How satisfied are you with the service that you get from your audiologist?	☐ Very unsatisfied ☐ Unsatisfied ☐ Satisfied ☐ Very satisfied	
42	Have you ever been fitted with hearing devices? (e.g. Hearing aids, cochlear implant)	☐ Yes ☐ No → go to 45	

43	How often do you wear hearing devices?	☐ 8 hours or more per day ☐ 4–8 hours per day ☐ 1–4 hours per day ☐ Occasionally (less than 1 hour per day but more than 1 hour per week) ☐ Rarely (less than 1 hour per month) ☐ Never	
44	How satisfied are you with your hearing devices?	☐ Very unsatisfied ☐ Unsatisfied ☐ Satisfied ☐ Very satisfied	
45	Do you typically take someone with you when you go to see the audiologist?	☐ Yes ☐ No → go to 47	

46	Which of these people do you take with you when you go to see the audiologist?	☐ Father/stepfather ☐ Father-in-law/Partner's father ☐ Mother/Stepmother ☐ Mother-in-law/Partner's Mother ☐ Spouse/De facto ☐ Partner ☐ Brother ☐ Sister ☐ Boyfriend/Girlfriend ☐ Own children/Stepchildren ☐ Other relatives ☐ Non-relatives	
47	Which of these people would you like to take with you when you go to see the audiologist?	☐ Father/stepfather ☐ Father-in-law/Partner's father ☐ Mother/Stepmother ☐ Mother-in-law/Partner's Mother ☐ Spouse/De facto ☐ Partner ☐ Brother ☐ Sister ☐ Boyfriend/Girlfriend ☐ Own children/Stepchildren ☐ Other relatives ☐ Non-relatives	

48	Do you get help with your hearing loss from any of these?	☐ Speech Pathologist ☐ Specialist Teaching Unit ☐ Teacher's Aide ☐ Teacher of the Deaf ☐ Educational Psychologist ☐ Other Psychologist ☐ Hearing Loss Support Group ☐ Social/Youth Worker ☐ Disability Employment Services Provider ☐ Occupational Therapist ☐ Speech Pathologist ☐ Specialist Teaching Unit ☐ Teacher's Aide ☐ Teacher of the Deaf ☐ Educational Psychologist ☐ Other Psychologist ☐ Hearing Loss Support Group ☐ Social/Youth Worker ☐ Disability Employment Services Provider ☐ Occupational Therapist	

49	When you go to see your audiologist, how important are these things to you?	Not important			Very important	
	My audiologist listens to me	☐	☐	☐	☐	
	My audiologist explains things clearly	☐	☐	☐	☐	
	My audiologist cares about me	☐	☐	☐	☐	
	I am involved in making decisions about my hearing loss	☐	☐	☐	☐	
	I trust my audiologist to make decisions that are in my best interests	☐	☐	☐	☐	
	I can see my audiologist whenever I need to	☐	☐	☐	☐	
	I see the same audiologist every time	☐	☐	☐	☐	
	My audiologist addresses my individual needs	☐	☐	☐	☐	
	My hearing aids work for me	☐	☐	☐	☐	
	I like wearing my hearing aids	☐	☐	☐	☐	

	My audiologist talks to me, not just my parents	☐	☐	☐	☐	
50	When you go to see your audiologist, how much do you agree with the following statements?	Strongly disagree			Strongly agree	
	My audiologist listens to me	☐	☐	☐	☐	
	My audiologist explains things clearly	☐	☐	☐	☐	
	My audiologist cares about me	☐	☐	☐	☐	
	I am involved in making decisions about my hearing loss	☐	☐	☐	☐	
	I trust my audiologist to make decisions that are in my best interests	☐	☐	☐	☐	
	I can see my audiologist whenever I need to	☐	☐	☐	☐	
	I see the same audiologist every time	☐	☐	☐	☐	
	My audiologist addresses my individual needs	☐	☐	☐	☐	
	My hearing aids work for me	☐	☐	☐	☐	

	I like wearing my hearing aids	☐	☐	☐	☐	
	My audiologist talks to me, not just my parents	☐	☐	☐	☐	
51	This question incorporated the MPOC-A in its entirety – please see Bamm et al. (2010) for details					

CHAPTER 5 - HIGH QUALITY AUDIOLOGICAL CARE ACCORDING TO YOUNG ADULTS WITH HEARING LOSS

Rationale

The eventual purpose of this thesis is to attempt to explore the experiences of hearing rehabilitation described by young adults living with hearing loss, so that hearing rehabilitation can be better tailored to this population. To that end, a subsample of the participants in the previous study were interviewed and their responses analysed, using a constructivist grounded theoretic methodology. The results of these interviews and analysis are presented in this chapter.

It is intended that a paper based on this chapter will be submitted to The International Journal of Audiology.

Introduction

The period of young adulthood (16–25 years of age) is one of great change for many young people, marked by transitions in a range of domains, including education, employment, social, and personal life areas. Educationally, many young adults move away from secondary schooling and into tertiary education; in employment, many enter the workforce for the first time or begin their careers; socially, many young adults move away from living in the family home and may even start families of their own; and personally, most begin to reject a dependence on others to make decisions in favour of a sense of autonomy (Arnett, 2000). This continual series of transitions is also associated with ongoing neurological and psychological development, culminating in the maturation of the prefrontal cortex and resulting stabilisation of decision-making ability in the twenties (Fuster, 2008, p. 17).

Hearing loss has been shown to have a range of impacts in these domains (Danermark, Granberg, Kramer, Selb, & Möller, 2013; Vas, Akeroyd, & Hall, 2017), and has

been associated with increased unemployment and lower overall life satisfaction in a survey of 113 young Australian adults with hearing loss (Chapter 4). This suggests that the transitions of young adulthood may be particularly impactful on those young adults living with hearing loss. In Australia, as of 2017, more than 8000 young people were receiving hearing services (including the provision of hearing devices such as hearing aids or cochlear implants) from the sole hearing service provider funded by the Australian Government to provide services to this age group (Australian Hearing, 2018, p. 4).

Among younger children, it is known that high-quality early intervention can significantly reduce the impacts of hearing loss on language and education outcomes (Yoshinaga-Itano, 2003) and that the educational benefits of intensive audiological (re)habilitation appear to persist into emerging adulthood, particularly in improving language skills (Goldblat & Pinto, 2017). However, a high proportion of young adults with hearing loss in Australia report not wearing hearing devices with which they have been fitted, and variability in satisfaction with hearing services is very high, suggesting that some young adults are not happy with the services that they are receiving (see Chapter 4)To date, there has been little research into why this variability may exist.

Part of the reason for this lack of investigation may be the limited understanding of what constitutes desirable or high-quality hearing care from the perspective of young adults with hearing loss. The incorporation of patient viewpoints and experiences into the design of care delivery models can assist services to achieve positive patient experiences, which are closely associated with improved patient safety and clinical outcomes across a range of health conditions (Doyle, Lennox, & Bell, 2013). As a result, partnerships between patients (and their families) and health professionals have been advocated as an important avenue towards improving the quality of health care delivery (American Academy of Family Physicians, American Academy of Pediatrics, American College of Physicians, & American Osteopathic

Association, 2007; Australian Commission on Safety and Quality in Health Care, 2011; Paparella, 2016). Primary to these partnerships is a shared understanding of how both parties define high-quality care so that efforts to improve care can be focused on achieving treatment goals that are tailored appropriately.

Importantly, shared understandings are highly context-specific, related to the disease process, the kind of care being delivered and to the patient population receiving that care (Hughes, Bamford, & May, 2008). As a result, efforts to improve care in a particular health care context need to be built on an understanding of the facets of care that are particularly important in that context. For example, fostering hope is an important factor when treating chronic health conditions, as the patient is likely to be living with the health condition for a long period (Hudon et al., 2012).

To date, while there has been no research addressing how young adults with hearing loss define high-quality hearing (re)habilitation, there have been efforts to understand the kinds of care valued by both young adult patients with other chronic health conditions and older adults with hearing loss, which may apply to this population. In a systematic review of the literature, we established three major facets of care that young adults with chronic disease value in their care: 1) emotional and social engagement with their practitioner, 2) empowerment of the young person and their family to make decisions and guide the health care process, and 3) an experience of care as effective at meeting their individual needs (Allen, Scarinci, & Hickson, 2018). Similarly, Grenness, Hickson, Laplante-Lévesque, and Davidson (2014) found in a qualitative interview study that a group of older adults with hearing loss particularly valued care in which they were kept informed of the relevant information, involved in the direction of their care, and in which their care was individualised to their lives and needs. While there are significant similarities between these two models, particularly the importance of care being tailored to the needs and desires of the patient, and

the incorporation of patients into shared decision-making, there are significant differences in emphasis. For example, while older adults with hearing loss want to be involved in care but left control of the encounters with the audiologist, young people living with chronic health conditions wanted to take control of the processes of care themselves. As a result, a simple "merging" of these two models is unlikely to produce a satisfactory understanding of high-quality audiological care among young adults living with hearing loss.

The aim of this research was therefore to explore the perceptions of what constitutes high-quality audiological (re)habilitation according to a group of young adult Australians with hearing loss. The intention was to enable audiologists and service managers working with this population to improve service delivery in ways that are likely to improve the experiences of the young adults accessing them.

Method

This study was conducted under the oversight of The University of Queensland Human Research Ethics Committee, the Australian Hearing Ethics Committee, and the Hear and Say Ethics Committee. Procedures were consistent with the NHMRC National Statement on Ethical Conduct in Human Research (National Health and Medical Research Council, Australian Research Council, & Australian Vice-Chancellors' Committee, 2007, Updated May 2015).

Research strategy

A Constructivist Grounded Theoretic methodology (Charmaz, 2006) was undertaken to understand how young people with hearing loss conceptualise positive hearing (re)habilitation. Grounded Theoretic methodologies allow researchers to generate theory to predict and explain human behaviour by exploring the understandings and reactions that underpin it (Glaser & Strauss, 1967). These methodologies utilise a set of methods, including concurrent data collection and analysis, theoretical sampling, and constant comparative

analysis, that distinguish them from other qualitative methods and make them particularly amenable to achieving rapid and highly targeted understandings (Birks & Mills, 2015). Constructivist Grounded Theory has at its centre the ideas of symbolic interactionism, in which research participants are active interpreters of their own experiences, decisions, and actions, who, in dialogue with a researcher, may construct a shared understanding of these (Charmaz, 2006, pp. 265-267).

The Grounded Theoretic approach was chosen due to its applicability to understanding areas of study that have not previously been explored, and where an understanding of experiences from the perspective of participants in the study is required (Birks & Mills, 2015). A Constructivist Grounded Theoretic approach was chosen in particular due to the clear explication of the methods involved (see Charmaz, 2006), as well as its congruence with the primary researcher's philosophical and epistemological approach.

Sampling

Initially, a theoretical sampling approach was intended for the selection of participants. Theoretical sampling of participants is an approach in which participants are selected based on their expected contribution to the emerging theory (Birks & Mills, 2015). In this sampling model, participants may be selected who have relevant experiences that may deepen existing understandings or potentially divergent viewpoints that may challenge emerging ideas. However, due to a very small participant pool, all participants who agreed to participate in this study were included. As a result, theoretical sampling of data was employed, in which the questions asked (and therefore the data sought and obtained) from them were modified as interviews progressed, to focus on elements of the emerging analysis (Charmaz, 2006). This method allowed the interviewer to challenge emerging areas of consensus by seeking out disconfirmatory examples from participants, as well as target data collection to facilitate more efficient theory generation.

Participants

Respondents to a previous survey study who were all young adult Australians living with hearing loss between the ages of 16 and 25 years were invited as part of that study to express interest in taking part in an interview study (see Chapter 4). The first author contacted all 33 participantswho expressed interest with further information about the nature of the interviews, and to make an appointment to conduct individual semi-structured in-depth interviews with those who then consented to take part.

Ten young people living with hearing loss participated in this study. They were aged 17 to 26 years, and all had been fitted with hearing devices (eight with hearing aids, one with cochlear implants, and one with both a hearing aid and a cochlear implant). All had bilateral hearing loss that was diagnosed during primary school or earlier. In the original survey from which they were recruited, three identified that they took their mother with them to audiology appointments, and four of the remaining seven reported that they would like to take their mother with them to audiology appointments.

All participants were of Caucasian Australian descent, and all used a spoken language at home. All had completed Year 12, except Imogen who was still in Year 11. Details of the participants are described in Table 5.1. Note that all names have been replaced by pseudonyms throughout this manuscript.

Table 5.1

Demographic details of participants

Participant	Age	Gender	Type of hearing loss	HA/CI	Home Language	Currently Studying	Employed	Living Situation	Time of diagnosis
Amelia	17	Female	Moderate Sensorineural	HA	English	Bachelor Degree	Part-time	Living with family	0–5 years
Brendan	24	Male	Moderate Sensorineural	HA	English	No	Full-time	Renting	0–5 years
Charlotte	18	Female	Moderate, type unknown	HA	English	Bachelor Degree	Part-time	Living with family	Primary school
Danielle	22	Female	Moderate Conductive	HA	French	No	Full-time	Renting	0–5 years
Elise	23	Female	Sensorineural, degree unknown	Both	English, some Auslan	Certificate III	No	Living with family	0–5 years
Felicity	22	Female	Moderate Sensorineural	HA	English	Postgraduate Degree	No	Living with family	0–5 years
Georgia	23	Female	Total, type unknown	CI	English	No	Full-time	Living with family	At birth
Henry	25	Male	Moderate Mixed	HA	English	No	Full-time	Living with family	Primary school
Imogen	17	Female	Severe, type unknown	HA	English	Year 11	Part-time	Living with family	Primary school
Julia	24	Female	Severe Sensorineural	HA	English	No	Full-time	Living with family	0–5 years

Note. Degree and type of hearing loss was self-reported.

Procedures

Eight of the interviews were conducted via teleconference using Zoom, in which a video-enabled chat room is created which participants then connect to using a mobile phone, tablet, or computer equipped with a webcam and microphone. Two of the participants elected to attend the University campus to be interviewed in person.

Table 5.2

Interview guide developed by researchers

Descriptive information about hearing history
Can you tell me about your hearing loss? • How would you describe your hearing loss? • How long have you had a hearing loss? • *Do you feel like your hearing loss gets in your way?*
Experience of audiologists
How long have you been seeing audiologists? Tell me about your experience with audiologists in managing your hearing. • How do you feel about your experiences with audiologists? • Tell me about some positive experiences you have had? • Tell me about some experiences which could have been different? How does going to the audiologist feel to you? • How have audiologists helped you in managing your hearing loss? In what way? • *What do you expect from your audiologist?* • *What characterises a professional audiologist?* • *What kinds of decisions do you make with your audiologist? Can you walk me through the process?* What sorts of things does your audiologist do that make you feel like they put you first?
Patient Centred Care in audiology
Tell me about your relationship with your audiologist? • What sorts of things would you feel comfortable telling them? • What sorts of things would you would not feel comfortable telling them? • How do you think your audiologist feels about trusting you to make decisions for yourself? Why do you think that?

- How do you feel about trusting your audiologist to make decisions that are in your best interests? Why do you think that?

What sorts of things do you do to manage your hearing outside of appointments?

- How does your audiologist assist you to do this? How do you manage these things on your own?

What sorts of things do you need from your hearing service that you don't think other people need?

- How does your audiologist help you with those things?
- In what ways is your hearing service specific to you? In what ways do you think that it's the same as what other people get?
- How does your hearing service work for you?

Who is in your family?

- How is your family involved in your hearing appointments?
- What sorts of things do you think your family should be involved in?
- What sorts of things should they not be involved in?

What sorts of things would make your hearing service more useful to you?

What kinds of things do you want to achieve with your life?

- How has your hearing affected what you have been able to achieve?

What would help you so your hearing didn't get in the way so much?

If you could change something about your hearing service, what would you change?

Note. Questions added over the course of data collection have been marked in italics.

An interview guide developed by the authors (see Table 5.2) was used in the individual semi-structured in-depth interviews. Individual interviewing allows participants to discuss and expand upon their experiences and views in their own words (Charmaz, 2006, pp. 85-86). All participants were given the option of having a support person present for the interview, and none requested that someone else be present, although due to the nature of the teleconference system, some participants did at times have a partner or family member present in the room during the interview. Interviews focused on the experience of hearing (re)habilitation, in particular how participants experienced their relationships with audiologists and hearing services, and how those relationships were developed over time. Each interview ended with an invitation to the participant to express anything that they wished to but had not yet, as well as to contribute further information by email should they

have any additional thoughts. One participant sent further comment by email, but the content did not relate to the present research question and was not included in the analysis.

Interviews were between 25 and 50 minutes in duration and occurred between June 2017 and May 2018. All interviews were recorded, transcribed verbatim by either the primary author or an external transcription service, and checked against the existing recordings and edited where discrepancies were found. These transcriptions were used for further analysis. Transcripts were analysed using NVivo 12.

Analysis

The methods of Constructivist Grounded Theory as described by Charmaz (2006) were used to analyse the interview transcripts, beginning after the first three interviews were conducted. In the initial coding phase, transcripts were read line-by-line by the first author, and all data related to the research question were identified and coded. Following this, emerging topics of interest were identified, and focused coding within these topics of interest was applied to those same transcripts to form categories. These emerging categories were then used for further focused coding of additional interview data. Focused coding is a process in which the data are coded specifically for aspects relating to a particular emergent category, to describe its content and delineate its boundaries. Focused coding particularly aims to elucidate the processes underlying and generating the data, rather than describing the data itself. Categories that appeared to be broadly applicable across the interviews and had explanatory power then became aspects of the theory.

As the theory emerged, it was then critically applied to the extant transcripts, to determine whether it adequately described and explained the data. Where disagreements between emerging parts of the theory and the data were identified, the emerging theory was modified to explain all the data, and the process was repeated.

A series of theoretical and interrogative memos were also kept by the first author, which identified areas for further theoretical sampling, theorised about interesting aspects of the data, and challenged areas of the emerging analysis. Some of these memos were ultimately incorporated into the results as presented below. Memos that identified potential areas of bias were discussed among the research team to enhance the trustworthiness of the findings.

Member checking was used to ensure that participants' experiences were accurately and appropriately represented in the results (Sandelowski, 2002). Once the initial analysis had been completed, the results were written up and sent to participants with an invitation for them to comment on the findings. Two participants replied, both endorsing the results, which have been presented in an expanded form below.

Researchers

Interviews were conducted by the first author, who also performed the initial coding. The first, second, and fourth authors were jointly responsible for all other tasks, and the third author provided review and commentary on the clinical applicability of the results during the production of the manuscript. The first author is an audiologist with five years of experience in (re)habilitative audiology in public practice in both urban and regional areas of Australia. He believes in and advocates for patient empowerment in hearing (re)habilitation and identifies with social constructivist and feminist approaches to research and clinical practice. This research forms part of his PhD studies. The second author is a speech pathologist and academic working in qualitative and quantitative research paradigms, with significant experience in working with families of clients with communication disability in both public and private practice. The third author is an audiologist working in a management role within the Australian Government-funded provider of (re)habilitative audiology services to young

adults. The fourth author is an audiologist and academic working in qualitative and quantitative research paradigms who has broad experience in hearing (re)habilitation.

Results

Participants described three primary pillars of quality hearing care: Getting the Basics Right, Being an Expert, and Giving Me the Power. Participants also described the importance of Developing a Relationship with their audiologists, building on these pillars. It should be noted that, in Australia, Government funding for (re)habilitative hearing services to young adults requires that services be provided by fully-accredited audiologists, rather than by audiometrists or other health professionals.

Getting the Basics Right

At the core of a positive experience for young adults with hearing loss is a hearing service that consistently addresses young people's individual needs. Valuable hearing interventions for young adults provide acceptable sound quality and a familiar hearing experience, with minimal risk of the experience becoming worse.

The young people in this study made special mention of audiologists who pay attention to their individual needs, rather than relying on assumptions about their hearing and communication needs. To be able to achieve this, young people expressed the need for audiologists to demonstrate an interest in the individual needs of patients, recognising that, due to the rapidly changing and diverse natures of young people, their needs and life contexts may be different to those of other young people whom audiologists might see. Young people emphasised the unique elements of their lives that set them apart from other young people, including attendance at boarding school, a passion for particular hobbies, or patterns of recreation with family. Recognising these differences was strongly identified as a sign of an audiologist delivering a high quality service. Audiologists who paid close attention to

individual needs were described as being better able to provide services (primarily hearing aids, cochlear implants, or other assistive devices) that consistently address them.

> *So I saw someone [audiologist] when I was in university, and they were asking how the issues with hearing aids in university and lecture style theatres were going, and then when I started working they asked about that and through that transition, and whether things needed to be adjusted because of that transition.* (Brendan)

Participants strongly valued the sound quality of devices that had been fine-tuned in collaboration with their audiologists over a long period and wanted a familiar hearing experience to continue. Their hearing experience was the product of a continual process of trialling new settings and devices, communication back to the audiologist for fine-tuning, and adaptation on the part of the young person. As a result, large changes to devices or settings were experienced negatively, as a potential threat to their quality of life. The process of adaptation was described quite separate from the activities of daily living, and as a significant impost on their ability to perform these activities.

> *I had to get used to the new ones [hearing aids], and I think it made it a little bit more difficult to iterate (sic) exactly what I wanted out of the new ones and to iterate (sic) that I actually was finding it quite difficult to adjust to them, and still am actually.* (Danielle)

In particular, this meant that participants preferred a conservative approach to their device management, where any changes were as minimal as possible (while still producing beneficial results) and easily rolled back. Being able to "go back" to previously acceptable settings or devices provided a strong sense of security for young people. Substantial changes to their hearing (re)habilitation were generally unwelcome and were avoided or delayed as long as possible.

[F]or a long time I relied on very old hearing aids, and then I got new ones and didn't wear them because I didn't like them and I preferred my old ones. (Danielle)

However, trials of new technologies or hearing devices helped them make decisions about the benefits, if any, of accepting change.

Felicity: *But not only was I offered them, I was offered the opportunity to try them, so.*

Interviewer: *So you could actually make a valuation yourself?*

Felicity: *Informed, yeah. I could make an informed decision.* (Felicity)

Importantly, young people recognised the drawbacks of "in-office" testing of settings or devices. They reflected on devices (such as FM systems) or settings often working well within the clinic, only to not work well in important social or educational situations.

I'd used an FM in school I think in year eight or year nine. I got an FM pack and it behaved really weirdly, it would work fine in the audiologist's office. I knew how to turn it on, I could copy them and then I'd take it to school, do exactly the same thing, and it wouldn't turn on. I took it home, wouldn't turn on. I took it to the office, it would start turning on and working fine. (Felicity)

Being an Expert

Young people expected that the audiologists and hearing services that they attended would have expertise in the various facets of hearing care: including but not limited to the technological aspects of (re)habilitation. Participants expected their audiologists to be able to demonstrate mastery of the technology that they were using, beyond being able to adjust and customise the device effectively.

The ability of the audiologist to apply technology in novel and creative ways to address the individual needs of the young person was more important to participants than the ability to get the "best" settings immediately. Importantly, young people wanted their

audiologists to have an open mind towards new ways to use technologies that may be beyond the technology's original purpose – the use of a sound-activated baby monitor to enable a young person to hear a smoke alarm, for instance:

> *So, I have some funding for assisted technology that I wanted to use for a smoke alarm that I can use in other people's houses. And my friend was saying that she uses something when she stays over at someone else's house, so that if smoke alarm goes off then she's aware of it. But it's like my audiologist didn't know. And then, when I found out exactly what my friend was talking about, it was a baby monitor sort of thing, so that it picks up the sound of a smoke alarm. And, I was like, "okay, now I know what this is that she's talking about!" and I say to my audiologist; "Well, this is what she's talking about. This baby monitor type alarm – but I noticed there's lots of different types, so I'm unsure which one would be the most suitable." And, my audiologist's first thing to say is, "you don't have a child, so how can I approve a baby alarm for you?"* (Elise)

Young people expected that hearing service staff should be comfortable with alternative strategies for effectively communicating with people with hearing loss, such as providing written information and the use of email, text messages, and Teletypewriter (TTY) instead of phone calls. Participants shared that they had observed reception staff at clinics raising their voices to other patients on the phone, which was reported as a jarring and unpleasant experience.

> *And I'm like "that's okay," give my number and I said "but can you message? When you call me, I'm not going to pick up." And they still call, they still call. Initially they started, they were sending appointment reminders […] via text messages. But they called me, "well your aid's ready" and left a voice mail and you could have messaged that or emailed that – why did you have to call? […] And when I'm in the actual*

room, you can hear receptionists, […] [talking] to someone who isn't hearing very well on the phone, but then they start yelling and start getting frustrated and roll their eyes and I guess the person on the other end can't see that they're rolling their eyes but, I wouldn't like that going on. (Elise)

Audiologists demonstrating openness and willingness to learn about the varied and potentially unexpected aspects of a young person's hearing experience was particularly important to the participants in this study. This understanding on behalf of the audiologist also demonstrated the audiologist's awareness of the young person's developing competency and control over their health care, allowing audiologists to support their emotional and audiological needs.

I always really like it when they make it clear that they understand like what you're after. Or they might understand. I think some audiologists were just shocked, for example, [that] I would want to hold onto my old hearing aids to 6 years when you can get new pairs every three years, and the technology's getting so much better et cetera, so it's always nice to have an audiologist that understands where you're coming from. (Danielle)

Participants recognised the importance of clinical records in the quality of care that they received, especially as they understood that continuity of care was difficult to obtain.

I went through a period of time there where I was consistently seeing a new audiologist and having to re-explain what the problem was. That was a little bit difficult, and I remember asking one day "why can't I just get the same audiologist?" and their response was basically "it's just the way it is, but you get booked in with who you get booked in [with]." (Julia)

Good clinical records were seen to be important in allowing all staff to be familiar with the patient's story and experience, and so that this information could be shared within

the organisation rather than being confined to a single audiologist or reception staff member. In particular, the young people noted that incorporating of the reasoning behind decisions made, the current life situation of the patient, and other psychosocial information were all seen as important elements of documentation to allow the future staff of the hearing service to recognise patterns that may emerge over time.

> *A lot of what you discuss, it doesn't all get written down, or they might write down notes but one person will write notes in a way that somebody else doesn't necessarily understand, or it doesn't pick up on the cues, or you write down dot points that will jog your memory – but you have to have context for those dot points to actually make sense.* (Felicity)

Finally, participants described the hearing (re)habilitation system as complex and often changing, and as such, they described a need for individual hearing services to be experts on the broader hearing healthcare and disability services system, reducing difficulties for the young person to access the services that they need. Being aware of external support and funding services and supporting or advocating for young people trying to access these services reduced friction and helped participants to gain access to services that they could benefit from.

> *Well, I'm getting slightly frustrated. I guess, some will say I'm trying to get some assisted technology with [national scheme]. And, [hearing service] said they are a provider and then I get told different information every single time I try to get something done. "Oh well, you know, you need to get clear approval." I'm like: "It's already there, I was not told that." And then the next time: "oh well, we can't order this for you because how can we justify this?" So ... You know, sometimes I feel like they're not really working with me.* (Elise)

Making young people aware of what is happening in the clinic room, reducing wait times, and ensuring that administrative information is kept up to date (such as contact phone numbers and addresses) were all thought by the participants in this study to reduce frustrations with the hearing service.

> *[An ordinary experience?] Getting a letter in the mail from my Grandma that's been forwarded from [capital city] to [capital city in a different state] because they still haven't saved our new postal address properly, saying that it's time to make the annual appointment.* (Felicity)

Giving Me the Power

Empowerment of young people to manage their care was seen as vital to a positive experience in audiological (re)habilitation. This principle was viewed as important throughout the healthcare journey, from the young person's initial decision to visit an audiologist through to decision making and ongoing (re)habilitation.

When taking case histories and building a list of issues that need addressing, participants saw it as important that audiologists be led by the patient, allowing them to define the issues that should be addressed by (re)habilitation. As discussed above, the issues that a young person feels are important may be very different to those that their peers may experience as important, and their needs may change rapidly, and so assumptions about what sorts of approaches are likely to be appropriate for a young person will likely be experienced negatively.

> *[The audiologist] keeps saying; "Oh, you could talk on the phone if you really try." And, I sort of explain that I don't see how that's really going to improve the issues I'm having […] I could probably talk on the phone with people's voices I know but, most people who want to contact me, who know me, won't call me.* (Elise)

Importantly, goal setting should be completed with the young person themselves, rather than with their parents or other carers, particularly once the young person is capable of making these goals themselves. Several of the participants described never remembering a time when the audiologist spoke to their parents rather than to them directly, and where this was the case these relationships were positive and productive.

> *She always talked to me. Whenever you see a clinician or a doctor or someone in the medical field, I feel like when I was a child they always just talked to the parent: "this is what your child has, this is how you deal with it," but whenever I would go to my audiologist, she would always talk to me. Instead of "these are the adjustments, are you okay with this?" she was always… She taught me to self-advocate and all of those skills, and I really appreciated that.* (Amelia)

It was also important to the participants that audiologists provide detailed and comprehensive information, covering all the practical (re)habilitation options. Comprehensive information was thought to facilitate informed decision making by the young person, allowing them to decide what direction they would like their care to take. Information should take multiple forms, including verbal and written information, and young people may benefit from experiential information gathered themselves through trials of technology or changes to settings.

> *I think maybe having a little ... I hate to call it a fact sheet, because I'm really not a fan of them ... but, even having a small resource online to say "hey, this is where you were, and this is where you are now, and this is how the change could possibly affect you."* (Julia)

> *But not only was I offered them, I was offered the opportunity to try them, so […] I could make an informed decision.* (Felicity)

Provision of information should not be at the expense of an informed clinical opinion; young people weigh the opinion of the audiologist highly and will use them as an important informational resource for decision-making.

> *It's a bit like if you're going to buy a new laptop: it is a bit overwhelming if you walk into the store and you just go "laptops! Which one do I choose? How do I choose? How am I supposed to know which one works for me?" If you just say "these are your options," how do I know what one is going to be reliable and whatnot?* (Felicity)

Young people discussed the process of making decisions about their (re)habilitation as one in which they weighed the evidence presented to them by the audiologist against the weight of years of experience living with the functional, emotional, and social implications of hearing loss and hearing (re)habilitation. The choice to reject or decline (re)habilitation options was described as a decision justified by a perceived lack of usefulness, a lack of expected use, or significant social or personal consequences. As a result, "doing nothing" is a valid option for (re)habilitation to take, and audiologists should respect the decision of young people not to act.

> *When I was offered the in ear tube things as opposed to the normal moulds, "you can try these ones, some people really prefer them because they are more discreet and you don't notice them," and I was able to try them, and I was able to find out that I hated them as they don't transmit the sound as well, so you can't hear as clearly, and then you need to crank up all the settings in your hearing aids to use them.* (Felicity)

> *I just said to him one day "I know you want me to try, but it's not so much that I can change it. It's just that the way that I do things doesn't make sense for me to change it."* (Julia)

Following treatment, outcome assessment should also be led by the patient, with their opinion of success or failure accepted and acted on appropriately by the audiologist. As

described above, young people are aware of the drawbacks of in-office testing, and so feeling welcome to return to the clinic if they continue to have difficulty is important.

> *They suggested "why don't we try this, and then if it doesn't work in a couple of weeks come back and we'll try something else." So for me at least they've pushed solutions to try but always with a flexibility of we can change it afterwards.* (Brendan)

Building the Relationship

These three pillars of care (i.e., Getting the Basics Right, Being an Expert, and Giving Me the Power) support the establishment of a strong therapeutic relationship between hearing services and young people. This therapeutic relationship is created and fostered at all stages during the young person's interactions with the hearing service, not only with the audiologist during scheduled appointments.

From the perspective of the young people in this study, in an ideal world, they would be recognised when they arrive at the clinic, be seen by staff who demonstrate an interest in all aspects of their life and who have access to file notes that accurate reflect their history, and participate in conversations that feel like they are entered into without a prior agenda.

> *Well it's real one on one conversation, they really go with what you're saying, more than any agenda they have.* (Charlotte)

Young people's ideal staff member would make time for patients (particularly outside of ordinary appointment time), think holistically about their needs, and tailor the hearing and (re)habilitative experience to the person in front of them. Through tailoring the experience in this way, audiologists and other hearing service staff could emphasise to their patients that they come first, and facilitate emotional supports that are highly valued.

> *My mom called her up one day and said, […] "There's something wrong. I don't know what it is, but she's saying that she's hearing something and can we get it fixed*

today?" And she said, "Yep." And it was on a Saturday or Sunday, and she said, "Yeah, come on over to my house. I'll fix it up for you." (Georgia)

Strong relationships between young people and their hearing services were thought to be built on trust and emotional support, particularly where the young person felt that their audiologist cared honestly about their feelings, and supported them to have hope for the future.

It's just smooth sailing. You walk out feeling "okay, I'm on top of this for now, and I'll see you in 12 months, and anything else I'll bring it up." (Julia)

It made me feel kind of special. It was nice to know someone cared and that they would help me if I was struggling with something. (Charlotte)

Sometimes, this came from staff members treating hearing loss as normal, something that can be lived with and managed, rather than as necessarily a source of stress and fear.

I always remember those who are enthusiastic, always keen to go "Okay, you have a problem, how are we going to fix it?" Coming from someone who's had a fair few gnarly problems in the last few years […] it's been really refreshing to get someone with the attitude of "yeah, okay, I know you're suffering a little bit, and we'll get to the bottom of it." […] you're the professional and I trust you to be able to help me, and this is why I come to you for advice on how I can improve or things that potentially are being a downfall for me at the moment. (Julia)

Where staff at a hearing service are warm and friendly, have the young person's best interests at heart, and empower the young person to manage their life independently, a strong relationship can be developed.

Discussion

This research aimed to explore the perceptions of what constitutes high-quality hearing (re)habilitation according to a group of young adult Australians with hearing loss. Three pillars of quality hearing care were described by participants: Getting the Basics Right, Being an Expert, and Giving Me the Power. These pillars supported young people to Develop a Relationship with their audiologist and the hearing service. The pillars of quality care described by participants in the current study are similar to several extant models of patient-centred care (PCC) and patient- and family-centred care (PFCC), including those developed in other populations undergoing hearing (re)habilitation within Australia.

The first pillar of care identified by participants, Getting the Basics Right, reflects the importance to young people of a hearing service that works, providing a hearing experience that supports their ability to function in everyday life. The importance of a service that addresses the patient's individual needs has been highlighted in models of PCC and PFCC, including those targeted towards young adults (Allen et al., 2018), those developed in audiology (Grenness et al., 2014), and general-purpose models aimed at being applicable across a range of diseases, patient populations, and service delivery systems (Epley, Summers, & Turnbull, 2010; Stewart et al., 2014). Participants in the present study strongly emphasised the importance of stability in their service, responding negatively to attempts to address new needs that might jeopardise a familiar hearing experience or reduce the effectiveness of hearing (re)habilitation with regards to currently-met needs. This aversion to risk rather than a belief in the potential of hearing (re)habilitation to solve emergent problems may reflect a lack of hope for the future, the fostering of which has been identified as an important aspect of patient-centred care for people living with chronic disease (Hudon et al., 2012). The importance of stability may also be reflected in several participants' expressed nervousness about ongoing access to hearing services as they age. A better understanding of

future-focus and hopefulness in this population, supported by future research, may assist clinicians in supporting young adult patients to understand what constitutes an acceptable level of risk when trying new technologies, settings, or strategies in hearing (re)habilitation.

In order to be able to support young adults to make changes to their hearing devices and (re)habilitation, young people stressed the importance of clinicians having a high level of skill with the technology (Being an Expert). A similar finding – that technical skills were assumed – was described in Grenness et al.'s (2014) description of patient-centred audiological rehabilitation for older adults with hearing loss, suggesting that the importance of skill at adjusting and predicting technology is related to hearing (re)habilitation itself, rather than to the age of the patients. Importantly, this is not highlighted in the same way in previous studies of PFCC among young people living with a range of chronic health conditions (Allen et al., 2018). This focus on clinicians' technological skill may be due to the emphasis within hearing service delivery on the use of healthcare devices, such as hearing aids and cochlear implants, rather than on drugs and behavioural interventions (Grenness, Hickson, Laplante-Lévesque, Meyer, & Davidson, 2015).

The technological focus of hearing (re)habilitation may also go some way towards explaining the importance for participants of familiarity of hearing experiences, including being able to "roll back" changes to devices or settings. With the increased utilisation of personal electronic devices such as smartphones and smartwatches and increased integration of these into specialised medical usage, healthcare devices are becoming more accessible, omnipresent, and self-manageable, encouraging young people to become more comfortable with them. Increased comfort with medical devices can lead to their incorporation into users' self-identity, which is not the case for medications or behavioural interventions (Matthewman, 2018, p. 25). As a result, changes to devices or settings may be experienced as

existential threats to portions of the sense of self, and so ensuring that they can be "rolled back" if necessary allows for the return of the self to familiar ground.

The third pillar of care, Giving Me the Power, is very similar to the factor *Empowering Patients and Families* previously reported in our model of PFCC for young adults with chronic disease (Allen et al., 2018). This focus on a high degree of transfer of power from the clinician to the patient is not highlighted in other models of patient-centred care, which highlight the importance of "finding common ground" (Stewart et al., 2014) or patient "involvement in" decision-making (Grenness et al., 2014). There is also a wide range in the degree of power over decision-making afforded patients and families reported in the family-centred care literature (Epley et al., 2010).

Similar to young people living with other chronic health conditions, the young people in the present study went well beyond wanting to be involved in their care or finding common ground with their clinician. Rather, they signalled a strong desire to drive care themselves, actively making decisions and holding the power of their own (re)habilitation. The difference in focus may be a consequence of the development of personal autonomy that is a feature of emerging adulthood (Arnett, 2006, p. 56). For young adults with hearing loss, communication (re)habilitation may represent an important opportunity for reclaiming decision-making from parents and other authority figures. While taking up decision-making may be beneficial to the young person's psychological development, increased autonomy in emerging adulthood has been associated with increased risk-taking behaviour, even where those risks are particularly pertinent due to the presence of a chronic health condition (Helgeson, Reynolds, Becker, Escobar, & Siminerio, 2014). Full disclosure of the risks and benefits associated with particular decisions about hearing (re)habilitation should be sought by clinicians to ensure that young people are able to make fully informed decisions, where this is legally permissible.

The centrality of a strong therapeutic relationship to the experience of (re)habilitative care is shared by the model of PCC in hearing rehabilitation of older adults developed by Grenness et al. (2014), and many models of family-centred care (Epley et al., 2010). In the present work, young people with hearing loss emphasised their relationships not only with the audiologist who sees them in the clinic room but with the other staff who make up the hearing service and the clinic itself. Participants strongly favoured a non-judgemental approach where they did not feel that staff with whom they interact have already decided the course of the conversation. Importantly, while direct communication with the clinician was an important facet of the relationship with the hearing service, and although participants desired relational continuity with audiologists, they believed that this was difficult to achieve. Instead, they spoke about the importance of continuity with the hearing service, particularly around the keeping and reading of high-quality, biopsychosocial clinical notes. The quality of the relationship with their service was reflected across the many and varied ways that they communicated with their hearing services, through letters, text messages, and visits to the clinic to obtain batteries or repairs for their hearing devices, and the frustration that can arise when the service attempts to communicate with them inappropriately, underscoring the importance of administrative staff to their experience of care.

The recognition of all staff as important to the experience of care is a central thread that runs through theoretical models of PFCC (Committee On Hospital Care & Institute For Patient- and Family-Centered Care, 2012; Stewart et al., 2014, pp. 315-317), as well as patient- and family-centred implementations of care delivery (Josephs & Brown, 2017). The results of this study underline the importance of all elements of communication between a young person with hearing loss and their hearing service to their experience of care. Practically speaking, these results suggest that training of clinicians alone is not sufficient to ensure that appropriately high-quality care is being delivered. Rather, all staff have an

important role to play in the development of a positive experience of care, and processes and methods of communication should be audited to ensure that they are contributing positively.

As has been discussed above, the model of care developed as part of this research is very similar to extant models of patient- and/or family-centredness, such as those proposed by Allen et al. (2018), Epley et al. (2010), Grenness et al. (2014), and Stewart et al. (2014). This similarity supports this model not just as a description of what young adult patients describe as desirable audiological (re)habilitation, but as a description of a kind of patient- and family-centred care than may be deployed by audiologists when treating young adults with hearing loss. Subject to the limitations of the study, which are discussed below, this model of care may now be appraised in practice, to determine whether its application leads to improved outcomes for young adults with hearing loss. While whether attempts to improve patient- and family-centredness in the care of young adults with chronic health conditions improves outcomes is currently an open question (Allen, Scarinci, & Hickson, in press), we have demonstrated that there is some evidence for improved life satisfaction with increased patient-centredness of care among young adults with hearing loss (see Chapter 4).

A limitation of the present study was that it was predominantly one of young women who wore hearing aids, with only two of the ten participants identifying as male, and one of these two participants unable to comment on many of the social aspects of care due to autism spectrum symptoms. This gender disparity is even more extensive than that seen in the survey from which participants were drawn (see Chapter 4). The study of the experiences of young men accessing hearing services should be prioritised to ensure that any gendered impacts of experience are not ignored. Also, recruitment for this study focused purely on young people who were attending hearing services (and who had a strong enough relationship with their provider to consent to be contacted for participation in research). Future studies on young people who are at risk of "dropping out" of hearing services should be prioritised, as these

young people are likely to have the strongest experiences of what constitutes both high quality and low quality hearing service delivery. Australia's unique situation in which a single organisation is Government-funded to provide services to this population could facilitate prospective studies to identify reasons why young people reject or avoid accessing hearing services.

These results suggest approaches that hearing services and individual clinicians can take in order to try and improve the quality of the services that they provide as experienced by their young adult patients. For example, hearing services may wish to improve administrative procedures to ensure that addresses and contact preferences are up-to-date and noted by all staff, train clinicians to recognise and accept patients' decisions to refuse technological interventions or to return to previous settings, and implement improvements in informational continuity, so that information provided by patients is not lost between clinical encounters. However, the effectiveness and clinical feasibility of these kinds of interventions have not yet been tested, which warrants further study.

In conclusion, findings of this study underline the importance of hearing services working closely with their young adult patients, allowing them to define the parameters of and drive their care. By providing a hearing service that addresses the expectations of young adults by getting the basics right, that demonstrates expertise in all areas of hearing service delivery, and that gives young people power throughout treatment, strong therapeutic relationships can be developed between the hearing service and their young person. These relationships can then form the foundation for collaborative hearing care on into the future.

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CHAPTER 6 – THE QUEST TO DIVEST: THE NARRATIVES OF MOTHERS OF YOUNG ADULTS WITH HEARING LOSS

Rationale

The purpose of this study was to explore the roles that family members take, and the roles that they and their young adult children feel that they should take, in hearing rehabilitation. To that end, a sample of family members were sought, nominated by the young adult participants described in Chapter 5, to be interviewed, with four mothers agreeing to be interviewed.

The first interview question asked of each mother was "Could you tell me a little about your family member's hearing loss?" This question elicited a story from each mother beginning at diagnosis and moving through their children's (re)habilitation to the present day. These stories were powerful and emotive, and pointed at an underlying narrative that had not been initially suspected by the research team. As a result, an analysis using a Narrative Inquiry approach, focused on the particular narrative described by each of these mothers, was undertaken. The resulting narrative is presented in this chapter.

It is intended a paper based on this chapter will be submitted to the International Journal of Audiology.

Introduction

"I can't really imagine taking anyone [to audiology appointments] besides my parents, because I don't think most other people around me would really understand." Danielle sighed, and looked away from the camera. *"I feel like they can articulate some of what I'm feeling, or some of what I'm experiencing, perhaps better than I can, because they have their perspective and they've seen it from day one. Whereas I don't think anyone else has that kind of visibility or understanding."* Like many young people with hearing loss, Danielle was diagnosed with a moderate, bilateral hearing loss as a young child, and so for

the first part of her life her hearing (re)habilitation was managed by her parents. An insightful and articulate 22-year-old working full-time after completing her Bachelor degree, Danielle had moved out of home already, and for the previous four years or so had been successfully managing her hearing (re)habilitation on her own, without her parents' direct involvement.

As Danielle's statement emphasises, family are important to hearing (re)habilitation, particularly among young children, where they are the primary deliverers of (re)habilitative services, and take a range of roles (Erbasi, Scarinci, Hickson, & Ching, 2018). However, for children with chronic health conditions such as hearing loss and diabetes that do not hinder their ability to self-manage, as they reach young adulthood, their parents can step back from direct involvement, allowing the child increased autonomy and control over their (re)habilitation. This withdrawal of parent-directed (re)habilitation can enhance the young person's ability to prepare for transitions in their care (Eaton et al., 2017). As a result, facilitating parents to relinquish control of the hearing (re)habilitation process to the child has the potential to empower young adults with chronic health conditions in general, and hearing loss in particular, to manage their own (re)habilitation.

It is unclear what factors help or hinder parents in their efforts to withdraw from their young adult children's health care. In a previous review of literature exploring how parents and young people define patient- and family-centred care, parents acknowledged that merely an intention to facilitate independence for their young adult children was not sufficient to guarantee that they were able to independently manage their healthcare (Allen, Scarinci, & Hickson, 2018). Rather, parents acknowledged that engaged practitioners whom they trusted to support young people to assume control of care were necessary for young people to achieve independence in their healthcare. However, there has been little published research to date exploring how parents develop this trust in practitioners.

For many young adults with hearing loss, this is further complicated as they leave home and move away from their parents, which presents a further barrier to parents attending audiological appointments with them (Allen, Scarinci, King, & Hickson, in preparation-b). In Australia, a large majority of young adults with hearing loss were diagnosed prior to entering primary school (Allen, Scarinci, King, & Hickson, in preparation-a), meaning that parents have been involved in their young adult children's hearing (re)habilitation for twenty years or more before the child assumes control over their hearing (re)habilitation. As a result, it is important to address parental experiences in a manner that recognises their long history and development.

Narrative approaches to patient and family experiences have been increasingly advocated as a way of understanding not only the current experiences and preferences of health care consumers but also the development of these experiences and preferences over time (Clandinin & Connelly, 2000). In a narrative approach, participants are able to tell their stories in a relatively unstructured fashion, with the analysis focusing on those aspects of the experience that are most important to the participant, rather than those aspects which might be most theoretically interesting or salient to the researcher (Patton, 2015, p. 130). After hearing the participants' stories, the researcher can then seek the underlying shared story common to the participants, which along with its social context is referred to as the *narrative*. By engaging with the narratives expressed by patients and families, healthcare providers can gain a better understanding of their values and motivations, facilitating their ability to tailor care (Wang & Geale, 2015). Improved understanding of the patient's narrative is particularly important where patients and their families have an ongoing experience of the health condition and its (re)habilitation. For these health care consumers, their personal experience of illness and care takes primacy over the medical narrative of care, cure, and remission (Frank, 1995, p. 6).

In his work "The Wounded Storyteller: Body, Illness, and Ethics", Arthur Frank described the experiences of the *remission society*, nested within society at large and made up of those who have largely recovered from an acute initiating condition but require continual ongoing monitoring and management (Frank, 1995). He described three major kinds of health narrative described by members of the remission society: *restitution*, in which medical intervention returns the person to where they were before; *chaos*, in which the experience of the health condition causes irreparable disruption to the patient's life and sense of self; and *quest*, in which the health condition becomes the impetus for personal growth and change (Frank, 1995).

The experience of ongoing engagement with health care is reflected in the lives of those who experience chronic health conditions and their family members, where continual self-management and ongoing interaction with health services is an ongoing necessity. Similar to the experiences of people living with an acquired health condition, diagnosis of hearing loss is known to be a traumatic event for parents (Kurtzer-White & Luterman, 2003), and hearing loss has a well-documented impact on family members (Scarinci, Worrall, & Hickson, 2012). Also, parents, particularly mothers, are the primary unit of hearing (re)habilitation for young children with hearing loss, meaning they "inherit" their children's ongoing interaction with hearing services (Erbasi et al., 2018). However, the third-party nature of these impacts makes it unclear what if any aspects of these narratives may also be lived out by parents of young people with hearing losses.

This study aimed to explore the narratives described by four mothers of young adults with hearing loss with particular regard to their involvement with their child's hearing (re)habilitation. The eventual purpose is to assist audiologists in understanding the journeys of mothers through audiological (re)habilitation of their young adult children, and to assist them in navigating those journeys.

Method

Research Approach

A Narrative Inquiry approach was used in this study. Narrative Inquiry highlights the ways in which people construct and retell stories about their lives, particularly how they give roles to themselves and others within those stories (Connelly & Clandinin, 1990). Importantly, Narrative Inquiry recognises that stories are not individual to a person, but are constructed relative to the social and cultural world surrounding the person (Clandinin & Connelly, 2000). The combination of both the story that a person is telling and an understanding of the sociocultural context in which that story was developed and retold is referred to as a narrative. Narrative Inquiry has been used as a method for understanding and describing patient experiences across a range of health disciplines, including nursing (Wang & Geale, 2015).

Sample/Data Collection

Ten young adult participants from a previous study (Allen et al., in preparation-b) were contacted and invited to nominate a family member of their choice who had experience of their hearing (re)habilitation and who would be willing to participate in an interview. Of these ten young adults, four responded and provided the details of their mother. Each mother was contacted and provided information about the study, an opportunity to ask any further questions, and to consent to take part. No fathers, siblings, or other family members were identified by the young adults as potentially relevant to the research.

Following receipt of informed consent, individual interviews were conducted by the first author (DA) with each participant using Zoom, a video-conferencing system which allows participants to speak in real time using a computer or mobile device with an internet connection and a webcam. Interviews were conducted between May 9th and May 28th, 2018,

and lasted between 20 and 30 minutes. The interviews aimed to explore the experiences of mothers of their children's audiological (re)habilitation. Each interview began with the question "Could you tell me about your daughter's hearing loss?" and then participants were free to shape the flow of the conversation as much as they wished. Each interview was recorded and transcribed verbatim using an external professional transcription service.

Ethical Approval

This study was conducted under the oversight of The University of Queensland Human Research Ethics Committee, the Australian Hearing Ethics Committee, and the Hear and Say Ethics Committee. Procedures were consistent with the NHMRC National Statement on Ethical Conduct in Human Research (National Health and Medical Research Council, Australian Research Council, & Australian Vice-Chancellors' Committee, 2007, Updated May 2015).

Analysis

An emergent analysis guided by the principles set out by Connelly and Clandinin (1990) was undertaken. Each interview transcript was initially coded by the first author to identify important events, experiences, and emotions expressed by the participants, and these codes were discussed among the research team. The four transcripts were then read in parallel, with particular attention paid to the major events and experiences described by each participant. Sections of each transcript relevant to the shared narrative were extracted and assembled into a single meta-story, which was then read repeatedly by the authors and interpreted. The authors interpreted the stories by engaging with them at the three levels of objectivation defined by Bourdieu (2004, p. 94): as reflections of the social environment that they shared with participants; by considering the participants' stories as descriptions of

clinical interactions; and in the context of the existing literature discussing parental experiences of having a child diagnosed with a health condition.

Trustworthiness

In order to validate the interpretation of the research team, a form of Synthesized Member Checking was used (Birt, Scott, Cavers, Campbell, & Walter, 2016). In this method, the data and interpretation are sent to participants, who are encouraged to provide feedback on the analysis. All participants were sent a preliminary version of the results appearing below, with an email invitation to respond, particularly on areas where they disagreed with the interpretation of the research team or felt that that interpretation was inadequate or misleading. Two participants acknowledged receipt, and one participant endorsed the interpretation and provided clarification of several of her quoted comments, which have been incorporated into the results presented below.

Researchers

All interviews were conducted by the first author, who also developed the narrative in consultation with the second and fourth authors. All authors were jointly responsible for all other tasks. The first author is an audiologist with five years of experience in (re)habilitative audiology in public practice in both urban and regional areas of Australia who believes in and advocates for patient empowerment in hearing (re)habilitation and identifies with social constructivist and feminist approaches to research and clinical practice. This research forms part of his PhD studies. The second author is a speech pathologist and academic working in qualitative and quantitative research paradigms, with significant experience in working with families of clients with communication disability in both public and private practice. The third author is an audiologist and academic working in qualitative and quantitative research paradigms who has broad experience in hearing (re)habilitation.

Results

Participants described their journey through their children's (re)habilitation over four stages: (1) Diagnosis: Guilt and Trauma, (2) Finding Strength in (Re)habilitation, (3) Becoming an Outsider, and (4) Receding into the Background. Within each stage, each relevant section of the meta-story is presented, followed by the associated analysis.

Diagnosis: Guilt and Trauma

> *She has had hearing loss for her whole life, I guess. But we didn't really realize that until she was probably about at kinder.* (Iris)

> *And back in the day, obviously they weren't tested for hearing before they came home, so we just merely took her home, first child, thought everything was fine.* (Janet)

> *We first started noticing something was wrong when she was about three years old. I talked to my friends about it, as you do, and they said, "oh, she probably needs grommets in her ears."* (Colleen)

> *As a mother, I knew there was something going on, but we couldn't put our finger on it.* (Anna)

> *It was a bit rocky to start with because we sort of felt really guilty that we hadn't realized there were problems with her hearing.* (Iris)

> *Took her in and […] I'll just never ever forget sitting there and the audiologist going "she has no fluid on her ear drum, so she doesn't need grommets." By then I realized that something major was going on because if she didn't need grommets, what was the cause of her hearing loss? I saw a specialist right then and there at the child health centre and the audiologist. I was pretty much told within the hour that my daughter would need aids for the rest of her life. I was just ... I was shocked.* (Colleen)

I must admit it was a bit numbing, I think when we first found out she couldn't hear, and because we had no one in the family that wore hearing aids or had any hearing problems, we were going blind, absolutely blind. (Janet)

When these four mothers thought about their daughters' hearing losses, every one of them began their story with their experience of the diagnosis. As their children were diagnosed before the introduction of universal newborn hearing screening, three of the four children were not diagnosed with a hearing loss until they were approximately three years of age. The delay in diagnosis lead to additional guilt for these mothers, due to the "lost time" in early childhood during which the children were without adequate hearing, which can have impacts on speech and language development.

For most of the mothers, this process began with a feeling that something was wrong, potentially in comparison to an older sibling or the child of a peer, and it was this feeling that prompted them to seek testing and eventual diagnosis. The realisation that something was wrong was described as a trauma by the mothers – and although all of them experienced it, they did so alone, without access to networks of other mothers who had previously dealt with the trauma of diagnosis. Where participants talked about other mothers, they described them as having a "normal" motherhood experience that contrasted with their own traumatic experience, marked by hearing loss diagnosis. In particular, other mothers' understandings of hearing loss diagnosis were described as more "normal": usually the diagnosis of a transient, conductive hearing loss due to middle ear effusion (a common condition during childhood). As a result, the transition from suspecting a transient hearing loss to the diagnosis of a permanent hearing loss marked the beginning of a different parenting journey from that of their peers.

This unique parenting journey was one in which they did not have any referents. None of the participants described having any friends who had children who had a permanent

hearing loss (likely due to the low incidence of permanent childhood hearing loss in Australia), and like most parents of children with hearing loss did not have experience of permanent hearing loss in their families, other than potentially ageing parents or relatives.

> *She was never sick. There was no warning that something like this would happen. I just assumed it was grommets. I hadn't even entertained any other possibility. Just to be told that, point blank with no preparation time … I mean, how do you ever prepare for something like that? I was devastated*. (Colleen)

> *Honestly for probably three days, I cried. Because it was just, "How is the world going to treat her?"* (Anna)

> *She's going to need to be aided for the rest of her life. I mean that's just blow upon blow. They did it as nicely as they could, […] but that's their world. They tell people this every day. It's not earth shattering to them. But I'm the parent, it's earth shattering to me. […] I remember when I heard the news one of the images I had in my mind, I don't know how it popped up, was her walking down the aisle with these big hearing aids in her ear. It's just not what you think of for your children.* (Colleen)

> *For me it was, it is what it is. [Her hearing loss] wasn't the end of the world. It was just, "How is she going to be when she grows up, and how is it going to affect her growing up in relationships?"* (Anna)

Diagnosis occurred in a diagnostic or hospital setting rather than the (re)habilitative setting with which they would eventually become more familiar. Importantly, the mothers reflected that this setting did not allow space or time for parental emotion, particularly grief and guilt. Grief and guilt are a feature of the maternal experience of having a child born with hearing loss, as with other congenital chronic health conditions (Kurtzer-White & Luterman, 2003; Landsman, 1998). The grief was described not only as grief for their children's past,

but for their future – their need to wear hearing aids, their future social standing, or the impact that hearing loss could have on their communication development and formation of relationships.

Anna and Colleen, in particular, described their diagnosis experience as particularly traumatic, provoking a strong emotional reaction. Of the four participants, these were also the two participants who were the most fearful about the future, particularly as their daughters grew older and "aged out" of the hearing system that they had become accustomed to since they began (re)habilitation as young children. The focus on the fear and uncertainty of this story of trauma may be a reflection of their current fears and concerns about the future. This is particularly interesting as both of these mothers identified fears about the future as the primary fear and trauma of their early experience following diagnosis.

Finding Strength in (Re)habilitation

> *My husband was in a bit of denial about it at first and I still remember I went in with him and met with an audiologist. […] She compassionately but very firmly said to my husband, "you're in denial and that's not going to do your daughter any good. We need to move forward here." She explained everything clearly. She was the person I would have liked to have been my first point of call rather than the child health centre. […] Rather than them doing the hearing test there and delivering the blow there.* (Colleen)

> *It's been a very strong relationship that we've had over, obviously, a number of years now, 13 or so years.* (Iris)

> *She was my daughter's audiologist […] at least eight to ten years. Continuity of care, I know it's not always possible, but it's so important. I remember just a little thing, whenever she had to be fitted for new moulds they'd pump that gunk in her ear. She*

hated that! She's such a brave girl, she never bucked up at anything but every time it was time for that, she hated it. [audiologist] knew it so she worked with her really well. Whereas when [audiologist] left we kept getting ones in who were all lovely, but they didn't know her. Not the way [audiologist] did. We were really blessed to have had her for as many years as we did. (Colleen)

We had [audiologist 1] to start with, and she was fabulous. She was actually instrumental in making sure that she managed her hearing aids and all that sort of stuff. So, we still don't know how to change the batteries and all that sort of stuff because she's done it all. (Iris)

I've been really pleased with them. They talk to her rather than to me. Even from when she was really little, which I found really impressive because it's about her, it's not about me. (Anna)

She was starting to self-manage her own hearing appointments from a very, very young age. (Janet)

She's been able to advocate for herself […] "These are the hearing aids, this is what we're gonna do, these are the options available to us, how would you like to go?" Well, it was always my daughter's decision about how she wanted to manage that. It's been fantastic really. (Iris)

The next major event described by respondents was the transition to (re)habilitative hearing services. In Australia, a sole Government-owned hearing service is funded to provide (re)habilitative hearing services to babies and children with hearing loss, and so each of the children was then referred by the diagnostic service to a local branch of this one hearing services provider. However, for mothers, the service was initially relatively immaterial –

rather, the audiologist that they were assigned became their primary point of contact and the primary support for managing and dealing with their child's hearing loss.

The (re)habilitative service was contrasted directly with the diagnostic service. While diagnosis had been a traumatic experience without space for emotion, the (re)habilitation journey began with a relationship with a particular person. This relationship with the audiologist was described by all four mothers very positively, setting up high expectations for hearing services going forward. In several cases, this first relationship was one that lasted for an extended period, contrasting with later experiences of continual change (described from the perspective of their children in Chapter 5). Continuity of care as a facilitator of trust in the audiologist and the hearing service was a recurrent theme at this point of the narrative.

The primary focus for all mothers was the way that, from very early in the process, the audiologist focused on empowering their child to manage their hearing loss and hearing devices, speaking directly to the child and allowing the parents to let go of control of the process. The concept that hearing service delivery should focus on the child's needs, desires, and capabilities, was repeated throughout each narrative as an important feature of the (re)habilitative journey – that it is the role of both the parent and the audiologist to empower their child to self-manage. The importance of empowering the child was identified not just as a responsibility of a parent of a child with hearing loss, but as a conscious choice of parenting style –each mother had independently decided to be the kind of parent who wants "resilient kids who can manage themselves" (Iris).

Becoming an Outsider

> *When you went to those appointments, you felt like you were a bit of an outsider. Because obviously, it was my daughter who was going there for her hearing aids and her moulds and things like that. And the audiologist would just purely talk to her or deal with her. It's only if they needed to ask a question or if she couldn't quite*

articulate what she was trying to say, they would then ask me. I felt like it was a little bit more of just someone on the sideline, just taking her to appointments. They didn't really ask me any questions, never asked how it was at home or how she was coping at school. (Janet)

When we first went and she first got her bone conductor, the audiologist was really adamant. I came over and said, "What do I have to do?" And she said, "Well you don't actually have to do anything. She needs to do this, this and that." So we were really supported very early on with an understanding that, yes, she needs to be able to manage it. (Iris)

All the support went to my daughter, I must admit. There wasn't probably any support for parents. If you had questions you just really had to ask questions yourself and I found that I just muddled my way along, like there was nothing really provided for parents and how to cope with it and where to go and what to do. (Janet)

Just to meet other kids and […] I would have liked to meet other parents and been able to talk to them about things. […] That would be something that I think would really be of assistance. (Colleen)

It's not that I'm not involved. I don't ... I wish for her to be happy and to be able to manage herself. And if she wants me more involved, I will. And if I'm not, if she doesn't want that, that's fine too. (Anna)

I'm not there to live her life for her. (Colleen)

The process of empowering their children was not universally pleasant or enjoyable for mothers. Rather, the focusing of care onto their children made mothers feel somewhat like an outsider in their own child's (re)habilitation. There resulted a tension between the mothers' desire to empower their child, their desire to ensure that appointments were as

effective as possible, and their desire to ensure that they had sufficient information and expertise to solve any problems that came along outside of appointment times. Parents felt that they were expected to allow their children to develop the skills that they would need in order to self-manage and that a necessary precondition for this was for them to temper their desire to "step in" and solve problems. This tension developed as the audiologist worked to transfer responsibility for managing interventions from the parent to the child.

This transfer of responsibility and the negative emotions associated with being removed from direct control of care challenged mothers' previous, almost unconditionally positive assessment of the audiologist as someone who understood the personal aspects of the hearing loss diagnosis and (re)habilitation process. However, perhaps due to the relationship that they had with the audiologist and the resultant trust that they had in them, this focus onto the child's ability to manage their care was accepted by parents as a necessary precondition for their children's development into functional, self-sufficient adults.

This focus on the children illuminated a perceived lack of support from audiologists for parents, both informationally and emotionally. All mothers identified a lack of support during the (re)habilitative journey, with a lack of understanding of the informational and emotional needs of the parents independent of the technical and skill-based needs of their children. This lack of maternal support was not presented alone, but always counterbalanced by their child's wellbeing: that is, mothers suggested that attention to the needs and desires of parents would necessarily come at the expense of the needs and empowerment of the child with hearing loss, and vice versa. For these mothers, the primary concern was always their children's development, and so they downplayed their own needs where they felt that addressing them would come at the expense of their children's independence.

Receding into the Background

She copes with it, but she doesn't really. I mean she does, but the older she's getting, the more she's articulating how hard it is and she's just had enough. (Anna)

It is hard to take a step back and ... She'll say now, because she's got new hearing aids at the moment ... It doesn't block out the background noise. Now, up until a year ago, her hearing aids have always blocked out the background noise. For whatever reason, they've changed, and she doesn't seem to understand why they've changed. She's struggling with it at the moment. (Janet)

Primary school was challenging. High school was as well, but primary school I think in many respects is more difficult, because kids just don't get it. And I think when you get older, your friendships develop a little bit more and she could ... She absolutely was still left out, that same sort of situation, but her core group of friends was better. They were more understanding and tolerant of her. Not tolerant, they were more accepting and happy to repeat things and understood when she didn't get it, she didn't get it. (Anna)

I'm there to support her and encourage her. (Colleen)

She's a young lady, she's a young adult. Comes a time where she's got to manage things on her own. (Janet)

My job as a parent is to prepare my kids to stand up on their own two feet, not to be dependent on me forever and her maturity and everything is at the point where I now feel she's ready. (Colleen)

I've never been one to control my kids. (Anna)

I always ask her, "How are you? How are you feeling? Look, you sound a bit nasally," or "Your speech is off." You can normally tell if something's wrong,

because she doesn't articulate as correctly as she can. She can get a little bit lazy in things like that. I'll often ask, "Look, are you okay?" or "Is anything upset? How are your hearing aids?" and whatnot. (Janet)

She knows, and they all know that when the wheels fall off, I'm always there. (Anna)

Does it ever go away? I don't think it ever goes away, Z. Seriously. My mum still says that now. I'm 50 years of age, and she goes, "You'll still be my baby." (Janet)

If something went wrong or she wasn't confident, or she had anything major that needed to be decided, we would support her in that obviously. (Iris)

If she wanted me to step in, absolutely I would. (Janet)

Each of the mothers who shared their stories was on very good terms with her daughter – all were still living with their daughters in the family home (which was true of the majority of the pool of young people who were a part of the precursor study) and so saw them regularly. However, none of the mothers was regularly or primarily involved in the child's hearing (re)habilitation, unless invited to for a specific reason, such as an appointment that was anticipated to be difficult or complex, or if a device was not working and the child's attempts to troubleshoot had failed. As a result, they described their roles as a loving third party who was well-placed to see the ongoing impact of hearing loss and the problems arising from hearing (re)habilitation.

For example, mothers were acutely aware of the ongoing social impacts of hearing loss, seeing continuing difficulties in social situations in the context of prior delayed social development. They also heard about problems with new or adjusted hearing aids directly from their children, particularly where adjustments had to be made to a new or altered sound signal. These stories caused them distress, highlighting the tension between their desire to empower their children (and to be the kinds of parents who produced independent and self-

sufficient children) and their desire to advocate and assist them in solving their problems. Having been the primary providers of advocacy for their children when they were young, the mothers in this study were aware of the power that they would be able to wield in hearing (re)habilitation, and that the drive to protect their children would not ever go away.

This tension, between the desire to empower their children and to assist them to solve problems, manifested in a number of ways. Some parents repeatedly checked in on their child's hearing and speech, others reminded them to discuss problems with their audiologists. All offered to attend appointments alongside their child – although always with the proviso that this was entirely the decision of the child, should they desire the parent's presence in the appointment. This reinforced mothers' sense of exclusion from the process, as the decision not to attend sparked feelings of not being "needed" by the child any more.

> *It's our parenting style. We want resilient kids who can manage themselves. (Iris)*
>
> *You do have to let your kids go at some point, and let them do it on their own. Been a good learning curve for me.* (Janet)
>
> *I'm not a babysitting kind of Mum. I try to enable my kids as much as I can.* (Colleen)
>
> *You want your kids to be happy and healthy and that's about it.* (Anna)
>
> *As most parents, you worry about your kids and you just wish that she would get what she needs.* (Janet)
>
> *Our role is to not helicopter parent her.* (Iris)
>
> *I just sort of sat to the side and listened to Charlotte and to her speak. I thought no, she's fine to do this.* (Colleen)
>
> *She's really sensible, makes wise choices. I feel that she will be fine with that and I'm happy for her to spread her wings, that's what kids do.* (Iris)

She knows it better than I do, and she's got a good relationship with the audiologist. (Anna)

She doesn't really rely on anybody else to do things for her. (Janet)

We really trust that she is really an old soul, she's got her head screwed on the right way. I'm very confident that things will go well. (Iris)

She'll find a way. Between us all we'll find a way. I just live by that. As in there's always a way to do whatever you want to do. You look at people who do amazing things and you echo that, "it's only your hearing. It's a big deal, but it's only your hearing. There's a lot of ways to get to where you wanna be." (Anna)

Each mother had a very clear idea of what kind of parent they wanted to be, and framed their decision about their child-rearing in the framework of this parenting style decision. All parents emphasised an authoritative style of parenting, in which they were responsive to their child's needs, but emphasised the child's abilities and responsibilities for their care. This awareness of their parenting style created a form of "conscious parenting" in which parenting decisions were filtered through an ideal of desirable parenting rather being based directly on the needs and desires of the parent. In this style of decision-making, the question of "what do I want as a parent?" was balanced against the competing questions of "what is best for my child in the long-term?" and "how would I behave if I was the kind of parent that I wish to be?"

The decision to step back from management of the child's care was supported by trust in both the child and their clinicians to act in a way that the parent felt was appropriate and in the child's best interests. In several cases, mothers talked about assessing interactions in an audiological appointment until they were satisfied with the relationship that their child and the audiologist had, allowing them comfort to take a step back. These assessments were

informed by the trust in their child that had developed over years of watching them develop into a young adult. However, it was also tempered by their experience of their child – not merely as a capable young person in the present, but also as a baby, as a child learning to function in the world, and as a future human being with potential that could be shaped by their health management.

In this way, the parent's decision to step back reflected the initial trauma of diagnosis and the resulting grief at the loss of their child's potentiality. The decision to finally release all control of their child's (re)habilitation was accompanied by a sense that, despite the additional challenges posed by hearing loss and ongoing (re)habilitation, their child would "find a way", with as much support from their family and clinicians as they felt that they needed.

Discussion and Clinical Implications

This study aimed to explore the narratives described by four mothers of young adults with hearing loss with particular regard to their involvement with their child's hearing (re)habilitation. The primary narrative expressed by these mothers began with their child's diagnosis, continued through the transition to a (re)habilitative audiology provider, and ended with mothers withdrawing from active participation in their child's care.

The narrative expressed by mothers may be seen as an example of Frank's quest narrative – one in which the person expressing the narrative uses the health condition as an impetus for personal development and change (Frank, 1995, p. 115). However, the quest of these mothers is fundamentally different from traditional quest narratives in several ways: as they do not have the hearing loss themselves, rather than a quest for them to overcome personal adversity, these mothers' goal was to remove themselves from (re)habilitation as much as possible. This quest is, therefore, a *Quest to Divest* themselves of power and responsibility for audiological (re)habilitation. Through this, the mothers hoped to empower

their child to self-manage, particularly in the face of the diverse problems that mothers envisage hearing loss may cause to their child's future. This quest is broadly aligned with the goals of patient-centred audiological practice as described elsewhere in the literature (Chapter 5; Grenness, Hickson, Laplante-Lévesque, & Davidson, 2014), in which patients are empowered to engage with decision-making and develop strong therapeutic relationships with audiologists.

The inclusion of parents and other family members in care is a central tenet of family-centred care (Epley, Summers, & Turnbull, 2010), and the involvement of family members in audiological rehabilitation is recommended across the lifespan (Scarinci, Meyer, Ekberg, & Hickson, 2013). However, mothers in this study reported a reluctance to speak up in appointments, which may complicate their engagement by audiologists and make the maintenance of their relationships with clinicians difficult. Audiologists should acknowledge that there are benefits in developing and maintaining a strong partnership with mothers as their children age, as mothers and audiologists have similar goals for young people with hearing loss. Early, long-term shared goal-setting may be useful as a way to manage maternal expectations and assist them in maintaining a feeling of inclusion in their child's life.

The recurrent pain resulting from the trauma of diagnosis is a feature of *chronic sorrow*, originally described in the 1960s among mother of children diagnosed with severe cognitive impairment (Burke, Hainsworth, Eakes, & Lindgren, 1992). When faced with a traumatic experience, a person may experience an acute grief reaction, which ordinarily ends with an acceptance of the new normal that the traumatic experience has precipitated (Lindemann, 1963). Chronic sorrow, on the other hand, is characterised by an inability for an initial episode of grief (in this case, to the hearing loss diagnosis) to resolve due to the child's ongoing needs and the presence of the health condition service as "a constant reminder of loss" (Burke et al., 1992). As the initial grief reaction remains unresolved, the parent

experiences recurrent episodes of sorrow for the loss of normality both for themselves and for their child occasioned by the disability. Unlike a normal grief reaction, the experience of chronic sorrow does not impair the parent's ability to manage their child's ongoing health care needs but does have an ongoing impact on the parent's mental health (Burke et al., 1992).

While chronic sorrow is considered non-pathological, its impacts on the parent's wellbeing make it important to recognise and reduce where possible (Burke et al., 1992). The suggested treatment for chronic sorrow is to recognise and support the parent, acknowledging their emotions and incorporating them into the care process (Peterson, 2017). In this way, the use of patient- and family-centred care, in which patients and families are included and empowered (American Academy of Pediatrics, American Academy of Family Physicians, American College of Physicians, & Transitions Clinical Report Authoring Group, 2011), is likely to improve the experience of parents living with chronic sorrow due to their child's hearing loss. However, the inclusion of parents in hearing (re)habilitation is complicated by the drive described above to remove themselves from active participation in their child's care. This, in turn, may contribute to the cycle of sorrow: as the familiar dynamic in which the parent is pivotal to the delivery of hearing (re)habilitation is challenged, the parent becomes an outsider to the (re)habilitative relationship, and their child's pervasive difficulties with hearing loss and (re)habilitation serve as further reminders of the trauma of diagnosis.

In the current Australian context, hearing services are only guaranteed until a young person turns 26, although this is changing with the roll-out of a National Disability Insurance Scheme, a Government program that will guarantee access to hearing services for young people with permanent hearing loss causing severe impairment. The impending transition of hearing services as their child "ages out" was a source of anxiety for mothers in the present study, and contributed to re-traumatisation and the re-emergence of grief. The roll-out of the

national scheme did not ameliorate this anxiety, as uncertainty around the processes of the new scheme (and the risk that their child would, in fact, not be eligible after all) were also highlighted as significant sources of worry. In the clinic, therefore, it is vital to actively engage parents as much as possible, in order to continue to manage the parent's ongoing grief in the face of their withdrawal from active participation in their child's care.

This ongoing sorrow resulted from a diagnosis experience that was described as a negative, medicalised experience, in which bad news was not delivered sensitively. Recent research conducted within diagnostic audiology suggests that while audiologists prioritise information about the hearing loss when delivering the news of a young child's diagnosis, parents are most interested in discussing their emotional reactions to the diagnosis (Geal-Dor & Adelman, 2018), and this disconnect seems to be a source of trauma for parents. Sensitivity to the kinds of information that recipients of bad news require is a feature of structured protocols for delivering bad news, such as SPIKES (Baile et al., 2000) or ABCDE (Rabow & McPhee, 1999). These protocols describe steps that practitioners should take when breaking bad news, and encourage practitioners to prepare for the event, be aware of the patient's level of understanding and informational needs, give the patient the bad news, and then deal with patient and family reactions. Both are commonly used in medical settings, although both were developed in the area of breaking bad news about life-limiting illness. To date, these protocols have not been evaluated for their effectiveness or usefulness in the context of the diagnosis of hearing loss to children, and so further investigation and development of a specific protocol for this setting is warranted.

The initial trauma of diagnosis was exacerbated in these mothers' stories by the delay in diagnosis that featured in each of the stories – three of the four children had hearing losses that were identified when they were approximately three years old, after the critical period for speech and language learning (Hurford, 1991). Delays in diagnosis of other congenital or

early-childhood chronic diseases have been associated with increased parental guilt (Kharrazi & Kharrazi, 2005). Since their children were born, universal newborn hearing screening has become available to most babies born in Australia, and has led to increased early diagnosis of congenital hearing losses and therefore early treatment with hearing devices (Australian Hearing, 2018, p. 12). However, a significant number of children with hearing loss are fitted with hearing aids later than the age of three (Australian Hearing, 2018). It is unclear whether this delay in treatment is due to a delay in diagnosis, which may be more likely to produce the kinds of guilt described above, or due to the relatively mild nature of the hearing losses identified making treatment unnecessary until the child enters school.

Limitations and Future Directions

This study included four passionate, engaged mothers, who were all living with their young adult children and who had close, healthy, and friendly relationships with them. Due to the nature of recruitment for this study, participants only identified family members with whom they had a positive relationship and who desired to take part in the research, rather than those who they were not so close to or who did not feel that there was a benefit to the research and their participation in it. Also, no fathers or other family members were identified by young people. The role of fathers in hearing (re)habilitation has been little studied, even in traditional two-parent households, although there is some evidence that fathers have different, support roles relative to mothers (Erbasi et al., 2018). As a result, it is unclear whether the experiences of fathers will be different from those of mothers in ways that require different kinds of management by clinicians. Future investigation that more closely involves all family members of young people with hearing loss – including fathers, siblings, and partners – may help facilitate the inclusion of diverse family members in a young person's hearing (re)habilitation.

The results of this study necessarily represent the experiences of only the four mothers involved in this study, and care should be taken not to assume that these results apply to all parents of young adults with hearing loss, or even all mothers of young people with hearing loss. The concordance of the narrative described by the four mothers does support its potential applicability, but the highly local nature of hearing service delivery means that experiences in other countries (and therefore other service delivery models) may diverge from the experiences expressed here. Replication of this study in other contexts should be prioritised to explore how broadly applicable these results are to mothers of young adults with hearing loss.

The results of this study are strengthened by the use of Synthesized Member Checking, which, we believe, facilitated participant engagement with the results by presenting them in an accessible format, with the words of participants displayed alongside the research team's analysis. Participants were able to follow the meta-story being presented, much as one might read a play alongside a commentary. Although only one participant responded to the results in a substantive way, we believe that her in-depth engagement with them supports our decision to use this method of member checking, and we encourage the use of this method in further research of this type.

Conclusion

Mothers of young adult children with hearing loss are an important and valued part of their children's hearing (re)habilitation. However, they describe their journey as one that largely involves removing themselves from involvement in their young adult children's hearing (re)habilitation, even when this may be at odds with their own emotional and psychological needs. By being aware of the tension that mothers experience between their commitment to their children's needs and their own needs, clinicians can help to ensure that

mothers are involved where appropriate, and supported to let go of control of their young adult children's care.

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CHAPTER 7 – SUMMARY, IMPLICATIONS, LIMITATIONS, AND FUTURE DIRECTIONS

Summary

The studies described in this thesis take a number of approaches to defining patient- and family-centred care (PFCC) for young adults with hearing loss and their family members. As little literature could be found directly addressing this topic, this thesis first sought to define PFCC for young adults living with a broader range of chronic diseases and explore whether efforts to achieve PFCC are associated with improvements in the effectiveness of care for this population. A systematic review of the literature, reported in Chapter 2, defined a model of PFCC generated from existing studies on the experiences of young adults with chronic disease and their family members. This model defined three major factors that comprise PFCC for the study participants: *Emotional and Social Engagement*, *Patient and Family Empowerment*, and the provision of *Individually Effective Care*.

A subsequent mixed-methods systematic review and synthesis of the literature (Chapter 3) examined the effects of interventions to achieve PFCC (PFCIs) as defined in Chapter 2 on the effectiveness and desirability of care. This study found that PFCIs are likely to improve the self-determination/self-efficacy of young adults with chronic diseases, supporting the importance of moving towards PFCC in this population. Interestingly, the studies identified in both of these reviews focused largely on chronic health conditions that are associated with significant social stigma (e.g., anorexia nervosa, schizophrenia). Social stigma is also associated with hearing loss (A. B. Barker, Leighton, & Ferguson, 2017), and so the findings of this review may be particularly relevant to young people living with hearing loss.

While the studies reported in the previous chapters addressed young adults with a range of chronic health conditions, the purpose of this thesis was to explore the nature of

PFCC for young adults with hearing loss, and so subsequent studies focused on young adults with hearing loss and their family members. Chapter 4 described the demographics and activity statistics of a sample of young adult Australians with hearing loss, highlighting the similarities and differences between young adult Australians living with hearing loss and young adult Australians in general. Overall, few differences were observed, although young adults with hearing loss were observed to be at higher risk of unemployment and to have lower overall life satisfaction than their peers. Importantly, increased perceptions of patient-centredness of received audiological rehabilitation was significantly associated with higher overall life satisfaction, supporting the findings from Chapter 3 that PFCC can improve patient experiences of care.

Central to the concept of PFCC is an understanding and prioritisation of the needs and desires of patients and their families, and to this end a sample of 10 participants from Chapter 4 then participated in an additional qualitative interview study (described in Chapter 5) to elucidate what they described as desirable, high-quality audiological care. Three pillars of PFCC in audiological practice were identified by participants: (1) *Giving Me the Power*, (2) *Being an Expert*, and (3) *Getting the Basics Right*. These aspects of PFCC facilitated clinicians and patients *Building the Relationship*, which was the eventual goal of high-quality care. Importantly, both this three-pillar model and the model of PFCC described in Chapter 2 were similar to existing models of patient-centred care, family-centred care, and PFCC that have been described in the literature (e.g. Epley, Summers, & Turnbull, 2010; Grenness, Hickson, Laplante-Lévesque, & Davidson, 2014; Stewart et al., 2014).

Young adult participants in the qualitative interview study described in Chapter 5 made strong references to the important role of family members in their audiological care over their lifetime. Therefore, a further qualitative interview study was conducted with family members of some of these participants in an effort to combine the young adults' perspectives

on the roles that family members do and should play in audiological rehabilitation with those of their family members. All the young adults who volunteered a family member to participate in this final interview study selected their mothers as the most important family member in their ongoing audiological rehabilitation. A narrative approach was used to describe the stories of four mothers regarding their experiences of their children's hearing losses (described in Chapter 6). This narrative, the *Quest to Divest*, describes how the mothers began their journey with trauma and guilt at the diagnosis of their child's hearing loss, then finding a safe place within rehabilitative audiology, and then working to recede into the background of their child's hearing rehabilitation, even when this transition was difficult or painful.

In this final chapter of the thesis, the theoretical and clinical implications of this work will be discussed, particularly as they relate to understandings and implementations of PFCC in the audiological rehabilitation of young adults. Several limitations of the studies that form part of this thesis will also be outlined, with recommendations for areas of further research that will extend and develop this work.

Theoretical Implications

Due to the relative lack of existing research into the audiological (re)habilitation of emerging adults, the purpose of much of this thesis was to provide a suitable basis for investigation to continue. Over the course of this work, four theoretical concepts were developed, which may be useful for future researchers. The first two, discussed here as *Patient- and Family Centred Discourses*, and *Biomedical and Humanist Approaches to Audiological Care*, are applicable to (re)habilitative audiology in general, both in clinical and research contextx. The importance and applicability of *Qualitative Methods in Audiological Research* is also addressed, and a case is made for increased use of interpretative methods in audiological research. Finally, the unexpected relationship between *Parents in Hearing Loss*

and Time is discussed, which may be useful to both clinicians and researchers working with the parents of children and young adults diagnosed with hearing loss.

Patient- and Family-Centred Discourses

This thesis has presented a variety of models of PCC, FCC, and PFCC (hereafter in this section referred to collectively as P/FCC), developed in and for a variety of health care contexts. For example: Stewart et al.'s model of PCC in medicine (2014); Epley et al.'s model of FCC in early intervention practice (2010); Grenness et al.'s model of operationalised PCC in audiological rehabilitation of older adults (2014); and the models of PFCC developed in this thesis for emerging adults living with chronic health conditions (Chapter 2) and living with hearing loss (Chapter 5). Each of these models has significant similarities (although with differences in approach and emphasis), as has been discussed in Chapters 2 and 5.

One explanation for these similarities is that these models have emerged from a set of underlying beliefs about the roles of patients and practitioners in health care interactions, which in turn lead to shared language used to describe them. The combination of belief and language is referred to as a *discourse*. Critical analysis of the discourses present in these models of care may help researchers, clinicians, and patients recognise the assumptions that they bring into audiological interactions, and challenge them where they may be inappropriate (Taylor, 2004). In this section, the major similarities between these models will be discussed in the context of a review conducted by Pluut (2016), which identified three major discourses that were present in models of patient-centredness developed across a variety of aetiologies, patient groups, and service delivery models. Pluut's three discourses – C*aring for Patients*, *Empowering Patients* and *Being Responsive* – represent three different ways of thinking about the roles that patients take in health care interactions

Providing care that reduces symptoms and disability to allow the patient to regain control over their body is an important part of delivering health care, and is highlighted by the concepts of *Individually Effective Care* in Chapter 2, and *Getting the Basics Right*, and *Being an Expert* of Chapter 5. The importance of care that allows patient or family to reduce the impact of a health condition and return to a "normal" day-to-day life is also reflected in the ideas of *Legitimizing the Illness Experience* (Hudon et al., 2012), *Individualised Family Services* (Epley et al., 2010), and *Exploring the Disease and Illness Experience* (Stewart et al., 2014) seen in other models of P/FCC discussed above. This kind of care, while recognising the importance of the patient's individual experience of their disease, is underpinned by a biomedical model of health care, in which disease is conceptualised as an abnormality that robs a patient of control over their body and in which health practitioners provide the patient with interventions that reduce their symptom burden (Engel, 1977). This discourse of patient-centredness is described by Pluut as *Caring for Patients.*

Engel (1977), in a seminal piece of work which challenged the biomedical approach to healthcare, pointed out that framing patients as individuals in need of assistance and support can be damaging. These kinds of power dynamics, he argues, can reduce the patient-practitioner relationship to one of paternalistic caring and protection rather than empowerment of patients through the facilitation of their needs. This can then result in patients moving their focus from a healthcare system that they may not understand to their practitioner, who is trusted because their role is to care for them (Arbuthnott & Sharpe, 2009). Due to the presence of a trusted "carer", in the position of the health care practitioner, patients may be encouraged to "give up" on understanding, deferring to the clinician's expertise (Stewart et al., 2014, p. 133).

The results discussed in this thesis suggest that this "giving up" may be unlikely among emerging adults with chronic health conditions, as a desire to be given control to

make decisions and lead care was a feature of the models developed in this thesis. *Patient and Family Empowerment* (Chapter 2) and *Giving Me the Power* (Chapter 5) both reflect this approach, in which *Empowering Patients* (the name ascribed to this discourse by Pluut) is a key feature. This focus on facilitating patients' capabilities to contribute to their own care is also seen in the ideas of being *Involved* (Grenness et al., 2014), *Family Strengths* (Epley et al., 2010), and *Finding Common Ground Regarding Management* and *Incorporating Prevention and Health Promotion* (Stewart et al., 2014). In this discourse, the patient and practitioner are considered more as equals, with the patient able to understand information and make decisions for themselves (Pluut, 2016). In this discourse, the practitioner's role is that of an informational resource, facilitating the patient to make informed choices about their own health care. This allows practitioners to relinquish some of the responsibility associated with being "in control" of a patient's care and acknowledges patients as active participants who are able to make their own decisions about their disease. As patients accumulate information that they can apply in managing their own health, they reclaim a measure of control over the narrative of the healthcare interaction without necessarily having to acquire the expert technical and informational expertise of the clinician.

The development of a therapeutic relationship between patient and practitioner is highlighted in a range of models of P/FCC, variously described as *Emotional and Social Engagement* (Chapter 2), *Building the Relationship* (Chapter 5), the *Therapeutic Relationship* (Grenness et al., 2014), *Enhancing the Patient-Clinician Relationship* (Stewart et al., 2014), *Developing an Ongoing Partnership* (Hudon et al., 2012), and *Family-Professional Relationship* (Epley et al., 2010). These portions of P/FCC models highlight the complexity of patients' beliefs and views, their changeability over time, and the requirement for clinicians to adapt to these needs. The requirement for clinicians to be adaptable to patient needs and desires led Pluut to label this discourse *Being Responsive*. In this discourse, the

patient's needs are paramount and the practitioner is a facilitator of wellbeing, and the patient's ultimate power in determining the flow and focus of their care is highlighted. Young adults in the studies described in this thesis strongly promoted the use of this way of approaching hearing care, particularly as a way of addressing the uniqueness and mutability of their needs.

The three discourses defined by Pluut came from a review of patient-centred models of healthcare, and so did not include discussions of family as a target of healthcare interaction. The incorporation of family means that the definitions do need to be modified slightly then thinking about P/FCC. When *Caring for Patients*, the third-party disability experienced by the family is also important, and so caring for the family and reducing the impact on them needs to be prioritised. Similarly, rather than only *Empowering Patients*, models that include families focus on empowering families as well as patients to achieve their needs. However, when *Being Responsive*, the work presented in this thesis recognises that incorporation of more people into the healthcare interaction also increases the number of people that the clinician needs to be responsive to (see Chapter 5). As a result, the assumption that the needs, values, and preferences of the patient are central is challenged, as the practitioner balances the potentially competing needs, values, and preferences of family members as well.

That the discourses presented by Pluut (2016) seem to underlie the models of care presented in this thesis suggests that efforts to achieve P/FCC for young adults with hearing loss may be supported by understanding efforts to improve patient- and family-centredness in other health care contexts, even when disease processes, aetiologies, or treatment modalities may be different. However, the differences between these models mean that it is not enough to merely transplant approaches from other areas of healthcare into (re)habilitative audiology – rather, consideration of the underlying beliefs, roles, and values assumed by practitioners is

likely to be useful. The conceptualisations of health and disease contained within these three discourses lie along a continuum, from the *biomedical* approach that prioritises disease to a *humanist* approach that foregrounds the person. This continuum is discussed in the following section.

Table 7.1

Mapping between discourses of patient-centredness presented by Pluut (2016), the factors of patient- and family-centredness for young adults with chronic disease defined in Chapter 2, and the elements of quality care for young adults with hearing loss defined in Chapter 5.

Pluut (2016)	**Chapter 2**	**Chapter 5**	**Approach**
Caring for Patients	Individually Effective Care	Getting the Basics Right	Biomedical
		Being an Expert	
Empowering Patients	Patient and Family Empowerment	Giving Me the Power	
Being Responsive	Emotional and Social Engagement	Building the Relationship	Humanist

Biomedical and Humanist approaches to Audiological Care

Biomedical and humanist approaches to care define a spectrum of beliefs about the nature of disease and a patient's role in the disease experience. While both patients' and practitioners' beliefs are important to healthcare interactions, practitioners' underlying beliefs about the nature and importance of the patient's individual experience and opinions can feed into therapeutic behaviours that they then play out in the clinic, potentially affecting a large number of different patients. The divide between a person's underlying philosophy and beliefs and their resultant behaviour is a central idea of cognitive approaches to psychology, and it is useful to mark the distinction before discussing the application of models of care in practice (American Psychological Association, 2018).

The biomedical model of care is typified by the medical gaze described by Foucault (1973). This is a process by which the practitioner "subtracts" the person living with a health condition from consideration to leave only those aspects of the health condition that are independent of the person, which then becomes the diagnosis (Foucault, 1973). In this approach to disease, diagnosis, and treatment, the experiences of the person receiving care must be "neutralised" and removed from the process of diagnosis and treatment, to avoid making an "incorrect" diagnosis. As a result, this approach values practitioners by their ability to use their judgement to make a "correct" diagnosis and provide similarly "correct" treatment.

On the other hand, the rise of phenomenology and subsequent awareness of the importance and variability of personal experiences of disease has led to the rise of humanist approaches to care (Payne, 2011), including the kinds of patient-defined models of PFCC discussed throughout this thesis. Originally developed in psychology, humanist approaches focus their attention on the person experiencing the health condition and their rights, capabilities, and values, and are now being advocated in a range of social and health care disciplines (Payne, 2011). In the humanist approach, consideration of the disease process is subservient to the patient's priorities. Underlying the humanist model of care is a philosophy in which it does not make sense to say that patients or practitioners are more important. Rather, the knowledge, experience, skills, and capabilities of patients and practitioners are incommensurable: a comparison between them does not make sense. The theoretical knowledge regarding hearing loss and its progression held by an audiologist, for example, is fundamentally different to the experiential knowledge of living with hearing loss held by the patient, and so this approach holds that it does not make sense to talk about one "knowing more" than the other.

In practice, many practitioners and academics advocate positions that blend these two approaches, in which both approaches are considered useful in the goal of addressing patient needs and reducing disability. The biopsychosocial model advocated by the World Health Organisation (WHO) is one example, exemplified by the two best-known arms of the WHO family of international classifications: the International Statistical Classification of Diseases and Related Health Problems (ICD-10, 2018) and the International Classification of Functioning, Disability and Health (ICF, 2002). The ICD-10 is, at its heart, a codification of Foucault's *rational space of disease*: a list of diagnoses that may be observed once the medical gaze neutralises a patient's personal experience (Foucault, 1973, pp. 9-11). In contrast, the ICF attempts to quantify the health of the individual as experienced by that person, independent of any diagnosable condition that may have caused a reduction in their health (World Health Organisation, 2002)

In the present work, the models of care presented in Chapters 2 and 5 also blend both biomedical and humanist approaches to care: care of the young person with a chronic disease requires care that is effective at reducing symptomatology, but which also recognises their social and emotional presence in the health care interaction. This is reflected by the spread of different aspects of each model across the spectrum of belief (see Table 7.1). Young people with hearing loss desired clinicians who were able to behave in ways congruent with both approaches: they strongly desired audiologists who were technical experts, effectively providing hearing services that made listening easier (a biomedical outcome), while also valuing services that build strong interpersonal relationships with them on an ongoing basis (a humanist outcome).

While both the biomedical and humanist approaches to care can be seen in the models of care that patients and families desired, participants had strong negative reactions towards experiences of care that they felt were overly medicalised (see Chapter 2, Chapter 5, Chapter

6). This suggests that positive experiences of care are shaped in particular by the presence of humanist patterns of thought and behaviour. This may be explained in part by existing work on audiological rehabilitation among older adults: in a qualitative study exploring what older adults with hearing loss desired from audiologists, Grenness et al. (2014) found that while technical skills were expected, interpersonal skills were particularly valued. In other words, biomedical thinking is required to be an audiologist, but it is holistic thinking that makes a patient- and family-centred audiologist.

Resulting from the beliefs held by the practitioner are a set of associated health care behaviours, audiological examples of which are presented in Table 7.2, drawn from the stories of hearing care told by young adults with hearing loss and their families who participated in the studies discussed in Chapters 5 and 6. Behaviours associated with the biomedical model of care are largely manualisable, and may therefore be more able to be learned without reference to the practitioner's underlying belief system. It is these kinds of technical skills that form the basis of audiological training and certification, potentially for this reason (Audiology Australia, n.d.; British Academy of Audiology, 2014; Council for Clinical Certification, 2018). On the other hand, behaviours associated with the humanist model are more interpersonal and relational, and may be more difficult to teach without reference to the underlying belief model.

As patients and families expressed a desire to be involved and recognised interpersonally, (re)habilitative audiologists working with a biomedical belief model (while likely highly effective at diagnosing hearing loss and prescribing treatment) may struggle to recognise the patient in the (re)habilitation process (Erdman, 2014). On the other hand, clinicians who hold a humanist belief model are likely to be more effective at meeting both the needs and desires of their patients and their families, and this kind of approach should be fostered among trainee audiologists.

Table 7.2

Examples of biomedical and humanist beliefs and behaviours

	Biomedical	**Humanist**
Beliefs	Hearing loss is separable from the patient	Hearing loss is primarily a patient experience
	Patient is the recipient of audiological intervention	Patient is the primary actor within audiological intervention
	Practitioner is the provider of audiological intervention	Practitioner is a facilitator of audiological intervention
Behaviours	Assessment of hearing loss severity in objective terms (e.g. audiogram)	Assessment of functioning, disability, and contextual factors from the patient's perspective
	Assessment of functioning in objective terms (e.g. speech testing)	Identification of patient goals (e.g. COSI, Dillon, Birtles, & Lovegrove, 1999)
	Prescription of hearing devices with appropriate settings	Determination of appropriate management and self-management strategies (e.g. hearing aids, communication training for patient and family)
	Provision of hearing aids with appropriate amplification	Implementing management and self-management strategies
	Assessment of improvement in functioning (e.g. aided speech testing)	Assessment of achievement of goals

It should be noted that experiences of hearing care discussed in this thesis are exclusively described from the perspective of patients and their families, and so the investigation of practitioner beliefs and approaches represents an important avenue for potential further research in this field. Rich descriptions of patient and family experiences have been facilitated in this thesis through rigorous qualitative investigations, underscoring the importance of qualitative research to inform clinical practice in this area.

Qualitative Methods in Audiological Research

The findings presented in Chapters 5 and 6 of this thesis are based on the study of some 10 young people with hearing loss and 4 mothers using strongly interpretive qualitative methods, a small sample from the perspective of traditional quantitative inquiry. The position of qualitative research in evidence-based practice is contentious, with a widening divide between qualitative and quantitative paradigms, named by Denzin and Lincoln (2005) the *fractured future*. Indeed, currently available manuals or reviews of evidence-based practice in audiology do not address qualitative inquiry as a method of achieving a clinically relevant understanding that may enhance patient care (Wong & Hickson, 2012). This may be due to the very recent rise of PFCC as a focus of hearing research, which has led to the increasing use of qualitative techniques such as Conversation Analysis (e.g., Ekberg, Meyer, Scarinci, Grenness, & Hickson, 2015), Qualitative Content Analysis (e.g., Grenness et al., 2014), and Thematic Analysis (e.g., F. Barker, Mackenzie, & de Lusignan, 2016). However, many of these methods are primarily descriptive, although thematic analysis does incorporate research interpretation as an important element (Vaismoradi, Turunen, & Bondas, 2013).

The use of qualitative methods like grounded theory has been advocated and applied in audiological research for some years (Knudsen et al., 2012; Meston & Ng, 2012). However, more interpretive methods such as narrative inquiry have seen less application in the audiological literature, although they are gaining in popularity (e.g., Hall et al., 2018;

Pillay & Moonsamy, 2018; Wallhagen, 2010). The use of both Constructivist Grounded Theory and Narrative Inquiry as primary research methods in the studies that are contained in this thesis led to rich understandings of the complex ways in which young adults and mothers experience and understand audiological (re)habilitation. The richness of these understandings has the potential to inform clinical practice in ways that are more relevant to young adults and their families, some of which are discussed later in this chapter. The results discussed in Chapters 5 and 6 stand as evidence of the usefulness, applicability, and feasibility of interpretive qualitative methods in audiological research, as well as their importance for understanding not only the contours of PFCC but the complex tapestry of patient and family experiences.

Parents in Hearing Loss and Time

When this thesis work began, it was hoped to develop an understanding of the roles that family members of young adults with hearing loss take in hearing (re)habilitation, the roles that family members desired to take, and the roles that young adults wished their family members to take. The intention was to develop suggestions for audiologists of how to best include family in the ongoing hearing (re)habilitation of young adults. However, young adults' understandings of the benefits of family inclusion and the roles that family members should play within audiological (re)habilitation (Chapter 5) were very different from those expressed by mothers (Chapter 6).

In particular, young people expressed these opinions using a very different concept of time to their mothers. Young people described decisions regarding family inclusion as happening in the moment – making choices about inclusion or exclusion based on their needs and requirements at the time of the appointment (discussed somewhat in Chapter 5). On the other hand, mothers' overarching metaphor for their children's hearing (re)habilitation was one of a journey: beginning with diagnosis, they were on a quest to empower their children so

that they could assume management of their own hearing (re)habilitation (Chapter 6). While both young people and their mothers attended the same appointments, and were involved in the same process of hearing (re)habilitation, the way in which they told these stories, and the kinds of time scales that they used, have important implications for future research into the experience of hearing (re)habilitation in this population.

Health sociologists have determined that living with a chronic health condition changes the way in which a person perceives and experiences time. In particular, living with chronic illness brings a person's focus onto the present day, a kind of "living one day at a time" (Charmaz, 1991, p. 178). This finding has been a reasonably robust and persistent feature of the chronic disease experience (see Corbin, 2003; J. C. Richardson, Ong, & Sim, 2008). This "present-focus" was also described by young adults with hearing loss in Chapter 5 as part of their decision-making process, which was primarily based on their needs and capabilities at the present time. However, the young adult participants in the present study had lived with their hearing losses as long as they could remember (either since birth or early childhood), and so did not have the drive to "recapture the past" that is a feature of the acquired chronic disease experience (Charmaz, 1991, pp. 193-195). As a result, it is unclear whether existing understandings of how time perspectives are changed by the experience of acquiring and living with chronic illness are likely to be applicable to young people living with congenital chronic health conditions, or to those who do not remember the acquisition of their chronic health conditions.

In contrast to their children, the mothers studied as part of this work were highly focused on the trajectories of their children's lives, particularly on the desired endpoint of those trajectories: independent and self-sufficient adults. As a result, they made decisions based not on what their children were experiencing in the moment but on the kinds of children that they wanted to raise (and wanted to have raised, once those children were

independent adults). This future focus, termed the *press for success* by parenting author Alfie Kohn, has been identified as both a potential motivator towards child success and a potential danger, in which "the child's present is essentially mortgaged to the future" (Kohn, 2006, p. 75).

However, the effects of this kind of parental decision-making orientation on the trajectory and outcomes of audiological (re)habilitation have not been studied in the peer-reviewed literature, and this remains an opportunity for future research. In addition, the interaction between the parental focus on the future effects of decisions and the young person's focus on immediate costs and benefits of decisions has also not been studied.

Clinical Implications

The results of the studies described in this thesis give audiologists working with young people with hearing loss some direction as to how they might structure and manage health care. Importantly, although the work focuses on the experiences and opinions of young adults, the results have implications beginning at diagnosis and extending through to young adulthood. These clinical implications fall into five major areas: (1) Empowering Young People, (2) Building Relationships, (3) Delivering PFCC in the Clinic, (4) Reducing Parental Chronic Sorrow, and (5) Implications for Audiological Education.

Empowering Young People

The implementation of PFCC has important benefits for the self-efficacy and self-determination status of young adults living with chronic disease, as discussed in Chapter 3. While this effect has, to date, not been investigated in audiological practice, participants in both Chapter 2 and Chapter 5 emphasised patient and family empowerment as an important part of PFCC, underscoring the important role that audiologists have in increasing young people's involvement in the processes of audiological (re)habilitation. This empowerment is a feature of many models of PFCC, although the way in which this is delivered is highly

context-specific (Chapter 2, Stewart et al., 2014). Participants also acknowledged the importance of beginning this empowerment process early, with both young adults with hearing loss and their mothers speaking positively of audiologists who directed care towards the young person with hearing loss from an early age (Chapter 5, Chapter 6). In Australia, young people are mostly legally able to consent to medical treatment and control their health data at the age of 16 (Bowles, n.d.). However, much of the relevant law addresses how to manage adolescent and young adult patients who do not wish their parents' involvement in the process. Given the strong preferences expressed by participants in the studies described in this thesis in favour of parental involvement, it is recommended that audiologists engage with parents early to manage the transfer of control over health decisions and data to young people commensurate with their level of maturity.

Structured, collaborative goal setting is one way of achieving patient empowerment in healthcare (Stewart et al., 2014), and this was highlighted by young adult participants, in both Chapters 2 and 5. Collaborative goal setting is an important practice within a range of rehabilitative disciplines (Wade, 2009), although within audiological practice there is significant scope for it to be delivered more commonly and effectively (F. Barker et al., 2016). In the present work, young adults highlighted patient-led goal setting as an important way for audiologists to address their rapidly changing needs, particularly given the time between appointments and the lack of continuity of care (highlighted by participants in Chapter 5).

"Non-adherence" to technological treatment is, anecdotally, a common complaint among (re)habilitative audiologists working with adolescent and young adult patients. The decision to "do nothing" is recognised as a valid and evidence-supported option for hearing rehabilitation among older adults (Laplante-Lévesque, Hickson, & Worrall, 2010), although this has not previously been addressed among young adults with hearing loss. In interviews

discussed in Chapter 5, young adults with hearing loss not only expressed their right to refuse technology (or, indeed, any treatment) but also explicated their process for doing so, which was based on assessing the expected value of the technology to the young person, its expected use, the social and inconvenience costs associated with it, and the perceived societal cost of an under-utilised technology that could be more useful to a different patient. Importantly, these young adults assessed technologies using a wider range of factors than older adults, who are primarily focused on the perceived benefit and comfort (McCormack & Fortnum, 2013). Expressing understanding for the decision-making process and engaging with the young person's reasoning may assist audiologists to support their young adult patients to make better-informed choices, particularly where those choices are made based on incomplete or misunderstood information. It will also assist audiologists to support young people with hearing loss who choose to refuse or return technologies, as they can be more certain that these decisions are well-considered, rather than purely based on dislike for technology, resistance to change, or inadequate benefit.

Building Relationships

The importance of building strong therapeutic relationships is also a feature of many models of PFCC, including within audiology (Chapter 5; Grenness et al., 2014). Young people interviewed in Chapter 5 identified clinically important relationships not only with their audiologists but also with reception and other staff of the hearing service. The interaction between the young person with hearing loss and hearing service staff when they arrive at the clinic represents an important moment, as this relationship is often more stable than that with their audiologist. In addition, this represents an opportunity for administrative information to be updated, as out-of-date administrative information can be a powerful driver of disengagement and frustration, particularly as the young person is striving to assert themselves separate from their family.

Interestingly, young adults described that they desired audiologists who make time for their patients outside of standard appointment times (Chapter 5). While young adult patients may want this option, it may not practically or legally be appropriate for many clinicians to provide within the bounds of reimbursement structures, insurance, professional boundaries, and life balance, all of which need to be taken into account when determining the design of hearing services. This desire on the part of patients may reflect a desire to feel connected with their hearing services. By setting clear expectations for the time and connectivity that they are able to provide (such as a commitment to a specified period within which email enquiries will be answers), or by making strategic changes to their availability (one evening of "late night trading" a week, for example), hearing services may be able to sate this desire for more connectivity without sacrificing the wellbeing of their staff. By co-designing changes with patients, hearing services can ensure that any modifications are highly targeted to the needs of their patients, improving standards of care most efficiently (Australian Commission on Safety and Quality in Health Care, 2011).

A major barrier to the development of strong relationships with clinicians is the lack of relational continuity that is a feature of the (re)habilitative audiological experience for many young people (see Chapter 5). Three different kinds of continuity of care are described in the medical literature: *relational continuity*, in which a patient sees the same clinical staff on an ongoing basis; *informational continuity*, in which the maintenance of clinical information, particularly through clinical record-keeping, means that a patient's information follows them over the course of their (re)habilitation; and *management continuity*, in which the approach taken towards a patient's treatment is coherent over time and between health care encounters (Haggerty et al., 2003). Young adults with hearing loss described relational continuity as highly desirable, and helpful for the development of strong therapeutic relationships, but a long history of changing audiologists meant that they no longer expected

it. Rather, they stressed the importance of informational and management continuity as ways of ensuring that appropriate care is delivered even in the absence of an ongoing interpersonal relationship with an audiologist.

Relational continuity of care improves adherence to treatment, patient satisfaction, and disease outcomes (Rosser & Schultz, 2007), and a lack of relational continuity was associated with reductions in delivery of PFCC to young adults with hearing loss (Chapter 4), making this an important potential target for improvements of care. Relational continuity of care should be encouraged, including allowing young adults the opportunity to follow their clinicians to a new location where this is practicable and desired. However, improved relational continuity of care was not associated with improvements in overall life satisfaction (Chapter 4), meaning that continuity alone is not sufficient to achieve broader psychosocial benefits for young adults with hearing loss. Rather, continuity of care facilitated the delivery of PFCC in the clinic, and it was this that led to significant life satisfaction benefits.

Delivering PFCC in the Clinic

Increased levels of PFCC have significant positive effects for young adults with chronic health conditions across a range of disease aetiologies (Chapter 3). It is therefore very positive that young adult Australians with hearing loss report high levels of PFCC as measured by the MPOC-A, compared to other studies of audiological (re)habilitation, and that improved levels of PFCC were significantly associated with improved overall life satisfaction (Chapter 4). However, the significant disparities in employment access between young adults with hearing loss and their normally hearing peers discussed in Chapter 4 have important implications for the kinds of services delivered to these patients.

Historically, audiological (re)habilitation of young people has focused on improving access to education, beginning with enriched language access to enable speech and language development and moving through to access to mainstream schooling (Gregory, Knight,

McCracken, Powers, & Watson, 1998). This focus on education seems to have borne fruit, with young adults experiencing similar educational outcomes to their normally hearing peers (Chapter 4). However, the lower employment access and stability reported in Chapter 4 suggest that young adults with hearing loss find it more difficult to obtain and maintain permanent, full-time employment, which has important implications for their ongoing health and wellbeing (Ross & Mirowsky, 1995; van der Noordt, IJzelenberg, Droomers, & Proper, 2014). As a result, audiological intervention should target the additional needs of young people entering the workforce, which may involve new and unfamiliar hearing environments, or regular use of telephones or other hearing-based communication. Determining the work-related needs of young people and implementing appropriate interventions may require additional appointments, more flexibility in intervention types, or increased trials of technologies and other management strategies compared to those required to address the relatively structured communication typical of classroom environments.

Reducing Parental Chronic Sorrow

The narrative described by mothers of young people with hearing loss presented in Chapter 6 has important implications not only for audiologists working with young adults but also for those working in neonatal and childhood hearing services, providing those audiologists some suggestions for respecting and managing the roles that parents take in hearing (re)habilitation. These implications are both for the moment of diagnosis, and for the process of transitioning control of hearing care from the parent to the young person.

The moment of diagnosis, identified by mothers as the beginning of their audiological (re)habilitation journey, was a source of trauma that sparked the chronic sorrow that was a feature of their experience of their children's (re)habilitation. The way in which bad news is delivered has been explored in the biomedical research literature, particularly around conditions that have a high or certain risk of death. This has led to the creation of structured

protocols for delivering bad news, such as SPIKES (Baile et al., 2000) or ABCDE (Rabow & McPhee, 1999). These protocols detail a process for practitioners to use to break bad news: they should prepare to give the news, explore the patient's or family's existing understanding and informational needs, give the bad news, and engage with reactions or emotions that may arise. However, these protocols have not been tested in audiological settings to date, and so it is not known whether the use of protocols like these would reduce the initial trauma experienced by mothers, and therefore their ongoing experience of chronic sorrow.

The primary treatment advocated for chronic sorrow among parents is recognition and support (Peterson, 2017), however mothers in the present research described not feeling as though they were recognised in the transition of focus of audiological (re)habilitation to their children (Chapter 6). The use of a structured transition process for young people with hearing loss has the potential to more clearly define the role of parents in transition, assisting them to feel more recognised. Guidelines for transition of youth from paediatric to adult care developed in general health settings suggest that transition happen in a structured fashion, beginning by the time the young person is 14 years of age and completing after they turn 18, and that the needs, questions, and opinions of the parent be addressed and considered (American Academy of Pediatrics, American Academy of Family Physicians, American College of Physicians, & Transitions Clinical Report Authoring Group, 2011).

Evidence suggests that where transition programs are implemented for young adults with hearing loss, they are closely focused on near-term goals and outcomes, rather than addressing their long-term needs (Luft, 2014). The results presented in this thesis suggest that in Australia, while audiologists are effectively transferring the focus of care to young people and empowering them to establish control over their care, that this is happening in a largely unstructured fashion, and without the needs or perspectives of family members (particularly mothers) necessarily being taken into account. The development of structured processes for

the transition of audiological care away from parental control and into the hands of young people living with hearing loss should be prioritised, particularly ones that facilitate parental knowledge of the process and their ability to be involved, heard, and responded to appropriately.

Implications for Audiological Education

As discussed previously in this chapter, the criteria by which formal programs of education for audiologists are accredited focus largely on technical skills, such as hearing assessment, device fitting, and the application of verification procedures, rather than the development of soft skills, such as active listening, counselling, breaking bad news, and shared decision-making. These soft skills were described by participants in the studies described in previous chapters as highly valuable, and an important part of developing the strong therapeutic relationship that is at the centre of many models of PFCC. The development of these kinds of skills is also described by audiology students as a desirable part of audiology programs (Tai, Woodward-Kron, & Barr, 2018). However, audiological communication is reportedly falling short of the desires of patients (Grenness, Hickson, Laplante-Lévesque, Meyer, & Davidson, 2015). The disparity between patient and clinician desires and the communication that is being delivered in clinics highlights the importance of developing audiological education programs, both within universities and as part of continuing professional development, that focuses on these soft skills.

For example, courses on working with young people with hearing loss may be run by community organisations such as Hear For You (http://hearforyou.com.au), an Australian advocacy and support organisation for teenagers and young adults with hearing loss. In addition, existing academic works on the challenges facing emerging adults may also be of use to clinicians and other hearing service staff who wish to better understand the challenges facing their emerging adult patients (e.g. Arnett, 2006; Konstam, 2015). However, there is

still a need for structured programs of continuing professional development for audiologists to develop their skills in this area.

Limitations and Future Directions

A number of limitations can be identified in relation to the studies conducted in this thesis and these give rise to suggestions for future research. While several of these have been discussed within the relevant chapters, they are discussed here together to facilitate the development of structured program of further research that can build on the work as a whole.

In particular, the systematic review described in Chapter 3 identified a relatively small body of literature with highly variable results. The further exploration of the impacts of efforts to achieve PFCC in young adults with chronic health conditions should, therefore, be prioritised, to determine what if any effects and outcomes are expected from PFCC so that implementation efforts can be assessed. The determination and application of appropriate outcome measures is vital to any effort to implement change within a health care system (Sansoni, 2016), meaning that the wide variety of outcome measures reported in the literature is a barrier to these results being applied in practice. Standardised measures such as the Hearing Aid Users Questionnaire (HAUQ) for assessing patient satisfaction and device use (Dillon et al., 1999), the 36-Item Short Form Health Survey (SF-36) to measure health-related quality of life (Chia et al., 2007), and the Measure of Processes of Care for Adults (MPOC-A) as used in Chapter 4 to measure patient-centredness (Bamm, Rosenbaum, & Stratford, 2010) will result in results that can be more easily applied by clinicians and health services in the future.

The studies described in Chapters 4 and 5 both had a relatively small number of self-selected participants with hearing loss, and those participants were mostly female, which is not in line with the best available data for young adults fitted with hearing devices in the community (Australian Hearing, 2018). Participants also tended to be well-educated

compared to young adult Australians at large, although it is unclear whether this may be due to increased intervention encouraging persistence in education or due to self-selection bias. It is unclear what impact (if any) this bias may have had on the results. Therefore, exploration of the academic and employment achievements for potentially excluded groups of young adults with hearing loss (such as males and those who have not completed secondary education) is warranted. This is especially important as men are known to access and utilise health services less than their female peers (Wang, Hunt, Nazareth, Freemantle, & Petersen, 2013). A population-level survey of young adults with hearing loss is uniquely possible in Australia due to their centralised treatment through Australian Hearing, and the collection of these kinds of data through existing clinical services would facilitate a deeper understanding of the challenges that face this group in the Australian context.

The gender bias seen in the studies discussed in Chapters 4 and 5 was also seen in Chapter 6, with only mothers volunteering to take part. Despite the varied nature of households in which young people with hearing loss lived (Chapter 4), participants in the study discussed in Chapter 5 largely spoke about their mothers when asked about family members who were important to their hearing rehabilitation. A recent study of parental involvement in hearing care for young children suggested that mothers are more involved in hearing appointments, while fathers take a more supportive role (Erbasi, Scarinci, Hickson, & Ching, 2018). If this result holds into young adulthood, young people may see their mothers as more involved in their direct hearing care than any other family member. However, hearing loss can be expected to affect all members of a family, and so perspectives of other family members including fathers, siblings, and romantic partners should be prioritised in future investigations of family in this population.

Importantly, the recruitment for this study was largely through hearing service organisations, and so these studies did not access young people who may have "dropped out"

of (re)habilitation. This means that the model of PFCC developed in Chapter 5 is specific to young adults who remain in care, and it is questionable whether its application will be effective in retaining young people who might otherwise "drop out" of hearing (re)habilitation. Due to the expanding nature of newborn and preschool hearing screening in Australia, a large proportion of young children with hearing loss are seen by (re)habilitative services, and longitudinal monitoring of these children may help identify factors that may predict later-life nonadherence. Preliminary research looking retrospectively at case files of young adults who do not use hearing devices as prescribed could also help to identify characteristics that appear to be predictive of reluctance to use hearing devices, which can be targeted as part of this prospective work.

In addition to research aimed at addressing the limitations of these existing studies, there are several surprising or novel findings that open up avenues for future research. Firstly, during interviews conducted with both young adults and mothers, several participants expressed feelings of being isolated from other people in a similar situation, and expressed a desire to be connected with peers. Peer support has been demonstrated to positively affect the experience of people living with other chronic diseases such as diabetes and asthma, helping them to feel less isolated, more empowered, and more confident with health behaviours, and improving the effectiveness of health interventions (Embuldeniya et al., 2013; Fisher et al., 2017). To date, although there is a growing body of work studying parental support of children with hearing loss (Henderson, Johnson, & Moodie, 2014), no clear research on the effect of peer support, either for young people or for parents, on the audiological (re)habilitation of young adults has been conducted. The implementation of peer support programmes has the potential to ameliorate some of the isolation expressed by both parents and young people. This kind of support network could be implemented relatively simply

through existing social networking tools (e.g., Facebook), through collaboration between clinicians and young people.

The development of structured peer support programs both for young adults with hearing loss and their parents may also offer an opportunity for parents to gain some measure of recognition for their ongoing audiological worry about their children as they move away from attendance at appointments, reducing their sense of chronic sorrow. Mothers in the study discussed in Chapter 6 confirmed that they did not know of support programs for parents of young adults with hearing loss, and that where they did once have access to a support program that they lost access as their children "aged out". This study was however only conducted on four participants, all of whom were mothers, and so more parents, including fathers, should be engaged to determine whether a service like this is considered useful and likely to be used by parents.

In addition to service-level interventions for reducing parental distress, psychological factors have also previously been shown to contribute significantly to the experience of distress for parents of children living with chronic health conditions (Sairanen, Lappalainen, & Hiltunen, 2018). This raises the possibility that interventions traditionally used in psychological therapies may be useful for modifying or reducing distress for parents and other family members associated with a child's hearing loss. In particular, Sairanen et al. (2018) found that increased mindfulness skills and decreased cognitive fusion were associated with lower distress, suggesting that therapies targeted at increasing mindfulness and cognitive defusion, such as Acceptance and Commitment Therapy (ACT), are likely to be effective at reducing parental distress. ACT can be effectively delivered in the clinic by non-psychologists (T. Richardson, Bell, Bolderston, & Clarke, 2018), which suggests that it could be trialled in the clinic by appropriately trained and skilled audiologists as a tool for managing parental distress.

Conclusions

The transition from adolescence to young adulthood involves a complex series of changes that are complicated by the experience of a chronic health condition such as hearing loss. This thesis has delineated some of the theory that can inform ongoing studies of the delivery of chronic disease management in general and hearing (re)habilitation in particular to young adults. This thesis presents preliminary understandings of the complex nature of the experience of young adults with hearing loss and sets the stage for further investigations into this fascinating, inspiring, and exciting population.

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APPENDIX – ETHICS APPROVAL LETTERS

THE UNIVERSITY OF QUEENSLAND

Institutional Human Research Ethics Approval

Project Title:	Patient- and Family-Centred Care for Young Adults with Hearing Loss and their Family Members
Chief Investigator:	Mr David Allen
Supervisor:	
Co-Investigator(s):	Dr Nerina Scarinci, Prof Louise Hickson, Alison King
School(s):	School of Health and Rehabilitation Sciences
Approval Number:	2016001270
Granting Agency/Degree:	HEARing CRC
Duration:	30th September 2018

Comments/Conditions:

Expedited Review – Low Risk

Note: If this approval is for amendments to an already approved protocol for which a UQ Clinical Trials Protection/Insurance Form was originally submitted, then the researchers must directly notify the UQ Insurance Office of any changes to that Form and Participant Information Sheets & Consent Forms as a result of the amendments, before action.

Name of responsible Committee:
University of Queensland Human Research Ethics Committee A
This project complies with the provisions contained in the *National Statement on Ethical Conduct in Human Research* and complies with the regulations governing experimentation on humans.

Name of Ethics Committee representative:
Professor Emerita Gina Geffen
Chairperson
University of Queensland Human Research Ethics Committee A
Registration: EC00456

G. Geffen

25/10/2016

Signature _____ Date _____

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Research and Innovation

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17th August 2017

Mr David Allen,
Doctor of Philosophy Candidate,
School of Health and Rehabilitation Sciences,
The University of Queensland

Dear David,

LETTER OF AUTHORITY AND RECOGNITION

The Hear and Say Research and Ethical Advisory Committee has reviewed the project entitled "Patient- and Family-Centred Care for Young Adults with Hearing Loss and their Family Members" for its suitability for Hear and Say related research.

The Committee is satisfied with all aspects of the protocol and consents to the involvement of Hear and Say in the project, subject to the conditions listed in the submitted protocol and any additional notes listed below:

- This approval is for the period of one year and expires on 15th August 2018.
- The principal investigator/s are accountable for adherence to the protocol by all researchers involved in the project.
- All research must be conducted according to the submitted protocol and contractual agreements, and if changes are made without the authorisation of the Research and Ethical Advisory Committee, approval for the project will be withdrawn.
- Any adverse incidents arising during the course of the research must be reported immediately to the Hear and Say Research and Development Manager, Dr Wendy Arnott, whose contact details appear below.
- A final report is required no later than one month after the above expiry date and should outline the study's significance, key findings, any difficulties experienced, and all publications (including those submitted for publication) arising from the research. Where a study has not resulted in any publications, the final report should include a brief discussion of the perceived barriers to publication.
- Hear and Say is to be notified of any publications arising after the study's expiry date.

If you require any further information, please contact Dr Wendy Arnott, the Hear and Say Research and Development Manager on (07) 3850 2221 or by email wendy.arnott@hearandsay.com.au

Yours sincerely

Dr Jane Black
Chair, Research and Ethical Advisory Committee

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